

# World Journal of *Clinical Cases*

*World J Clin Cases* 2021 December 16; 9(35): 10746-11121



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The *WJCC* is now indexed in Science Citation Index Expanded (also known as SciSearch®), Journal Citation Reports/Science Edition, Scopus, PubMed, and PubMed Central. The 2021 Edition of Journal Citation Reports® cites the 2020 impact factor (IF) for *WJCC* as 1.337; IF without journal self cites: 1.301; 5-year IF: 1.742; Journal Citation Indicator: 0.33; Ranking: 119 among 169 journals in medicine, general and internal; and Quartile category: Q3. The *WJCC*'s CiteScore for 2020 is 0.8 and Scopus CiteScore rank 2020: General Medicine is 493/793.

**RESPONSIBLE EDITORS FOR THIS ISSUE**

Production Editor: *Jia-Hui Li*; Production Department Director: *Xiang Li*; Editorial Office Director: *Jin-Lei Wang*.

**NAME OF JOURNAL**

*World Journal of Clinical Cases*

**ISSN**

ISSN 2307-8960 (online)

**LAUNCH DATE**

April 16, 2013

**FREQUENCY**

Thrice Monthly

**EDITORS-IN-CHIEF**

Dennis A Bloomfield, Sandro Vento, Bao-Gan Peng

**EDITORIAL BOARD MEMBERS**

<https://www.wjgnet.com/2307-8960/editorialboard.htm>

**PUBLICATION DATE**

December 16, 2021

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<https://www.wjgnet.com/bpg/gerinfo/208>

**ARTICLE PROCESSING CHARGE**

<https://www.wjgnet.com/bpg/gerinfo/242>

**STEPS FOR SUBMITTING MANUSCRIPTS**

<https://www.wjgnet.com/bpg/GerInfo/239>

**ONLINE SUBMISSION**

<https://www.f6publishing.com>

## Spermatocytic tumor: A rare case report

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**Author contributions:** Hao ML was responsible for the data curation; Hao ML reviewed the literature and contributed to original manuscript drafting; Li CH was responsible for the review of the manuscript; all authors issued final approval for the version to be submitted.

**Informed consent statement:**

Informed consent was obtained from the patient for the publication of this case report.

**Conflict-of-interest statement:** The authors have nothing to disclose.

**CARE Checklist (2016) statement:**

The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

**Country/Territory of origin:** China

**Specialty type:** Andrology

**Provenance and peer review:**

Unsolicited article; Externally peer reviewed.

**Peer-review report's scientific quality classification**

Grade A (Excellent): 0  
Grade B (Very good): 0  
Grade C (Good): C

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### Abstract

#### BACKGROUND

Spermatocytic tumor is a rare, malignant neoplasm of the testes. Since the prognosis for this tumor type is favorable, accurate diagnosis and differentiation from other malignant testicular neoplasms (classic seminoma and lymphoma) are crucial. To add to the existing literature on the diagnosis of spermatocytic tumor, herein we report the detailed clinical and histopathologic findings for a case that we encountered.

#### CASE SUMMARY

A 60-year-old Chinese man presented with a solid mass in the right scrotum. The mass was surgically removed and spermatocytic tumor was diagnosed. On microscopy, the tumor cells displayed an unusual arrangement in lobules, presenting a pseudo-glandular appearance. To summarize and compare the diagnostic features of this tumor and those of the differential diagnoses, we report our case findings and those mentioned in the literature for various testicular tumors. Although imaging methods can detect masses early in development, their diagnostic capabilities are limited. Biopsy, histopathology, and immunohistochemistry are necessary for confirmatory diagnosis.

#### CONCLUSION

It is important to identify and review the key diagnostic features of spermatocytic tumor.

**Key Words:** Spermatocytic tumor; Germ cell tumor; Immunohistochemistry; Pseudo-glandular; Case report

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**Core Tip:** Spermatocytic tumor is a rare malignant testicular tumor. At present, there are few reported cases in the world. The understanding of clinicians and pathologists about

Grade D (Fair): 0

Grade E (Poor): 0

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**Received:** July 30, 2021**Peer-review started:** July 30, 2021**First decision:** September 1, 2021**Revised:** September 6, 2021**Accepted:** October 21, 2021**Article in press:** October 21, 2021**Published online:** December 16, 2021**P-Reviewer:** Exbrayat JM**S-Editor:** Yan JP**L-Editor:** Wang TQ**P-Editor:** Yan JP

this disease is not comprehensive enough. Here we report a case of spermatocytic tumor, emphasizing the morphological features of spermatocytic tumor and the key points of differentiation from classic seminoma and lymphoma, so as to avoid misdiagnosis.

**Citation:** Hao ML, Li CH. Spermatocytic tumor: A rare case report. *World J Clin Cases* 2021; 9(35): 11115-11121

**URL:** <https://www.wjgnet.com/2307-8960/full/v9/i35/11115.htm>

**DOI:** <https://dx.doi.org/10.12998/wjcc.v9.i35.11115>

## INTRODUCTION

Spermatocytic tumor is a rare malignancy that accounts for 0.61% of testicular germ cell tumors[1]. Tumor development is independent of ethnicity and a history of cryptorchidism. Older men with an average age of 52 years are often the most affected [2]. At present, as spermatocytic tumors are extremely rare, there is a paucity of data for clinicians and pathologists to make differential diagnosis. Herein, we present a case of spermatocytic tumor, and review and consult the relevant literature. We summarize the morphological characteristics of spermatocytoma, hoping to provide clinicians and pathologists with more reliable diagnostic data and diagnostic ideas to help them serve patients better, reduce patients' pain, and avoid unnecessary, if not harmful, treatments.

## CASE PRESENTATION

### Chief complaints

A 60-year-old Chinese male farmer presented with a 3-mo history of right scrotal enlargement.

### History of present illness

The patient presented with right scrotal enlargement and had no scrotal tenderness, chills, fever, or other discomfort. No abnormality of the left testis, epididymis, or spermatic cord was discerned. The patient did not self-medicate or seek alternative therapies. He reported no lumbar or abdominal pain and no increased frequency, urgency, or pain associated with urination, but had a slight weight loss.

### History of past illness

The patient had no history of trauma, tuberculosis, or other relevant infectious disease.

### Personal and family history

The patient denied any family history.

### Physical examination

A 4 cm × 5 cm, slightly moveable, solid mass was palpated in the right scrotum, which drooped and was pale in color. No normal testicular or epididymal structures were palpated in the affected testis. No abnormality of the left testis, epididymis, or spermatic cord was discerned.

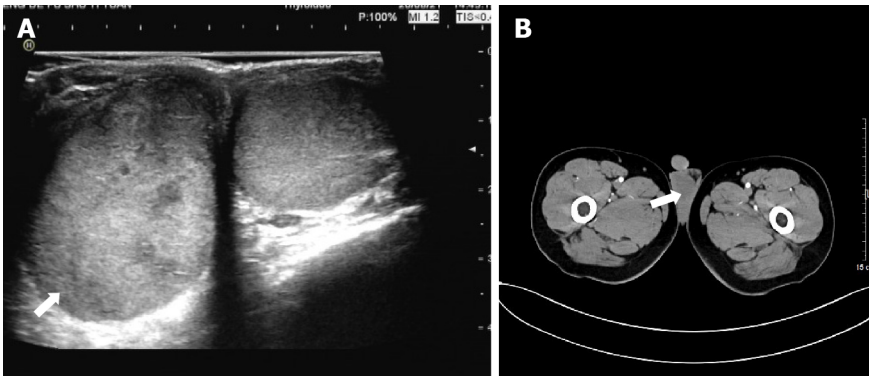
### Laboratory examinations

The results such as routine hematological testing, blood sedimentation rate, vascular endothelial growth factor, human chorionic gonadotropin (HCG), serum carbohydrate antigen (CA)199, CA125, CA153, alpha-fetoprotein (AFP), thymidine kinase 1, and carcinoembryonic antigen (CEA) were normal.

### Imaging examinations

Within 1 wk from presentation, the patient underwent scrotal ultrasound showing a 4.5 cm × 2.7 cm × 3.7 cm oval-shaped hypoechoic mass, with uneven internal





**Figure 1** Ultrasound (left) and computed tomographic scans (right) of the testicles. A: Increased volume of the right testicle, with a lesion of uneven parenchymal echo (white arrow); B: Uniform nodular change in the right testicle (white arrow).

echogenicity in the right testicle (Figure 1A). Color Doppler showed scattered color blood flow signals within the lesion. Meanwhile, the patient underwent computed tomography (CT), which revealed an enlarged right testis, with indistinct contour, uneven density, and uniform nodular change (Figure 1B). No normal testicular or epididymal structures were palpated in the affected testis.

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## FINAL DIAGNOSIS

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Spermatocytic tumor.

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## TREATMENT

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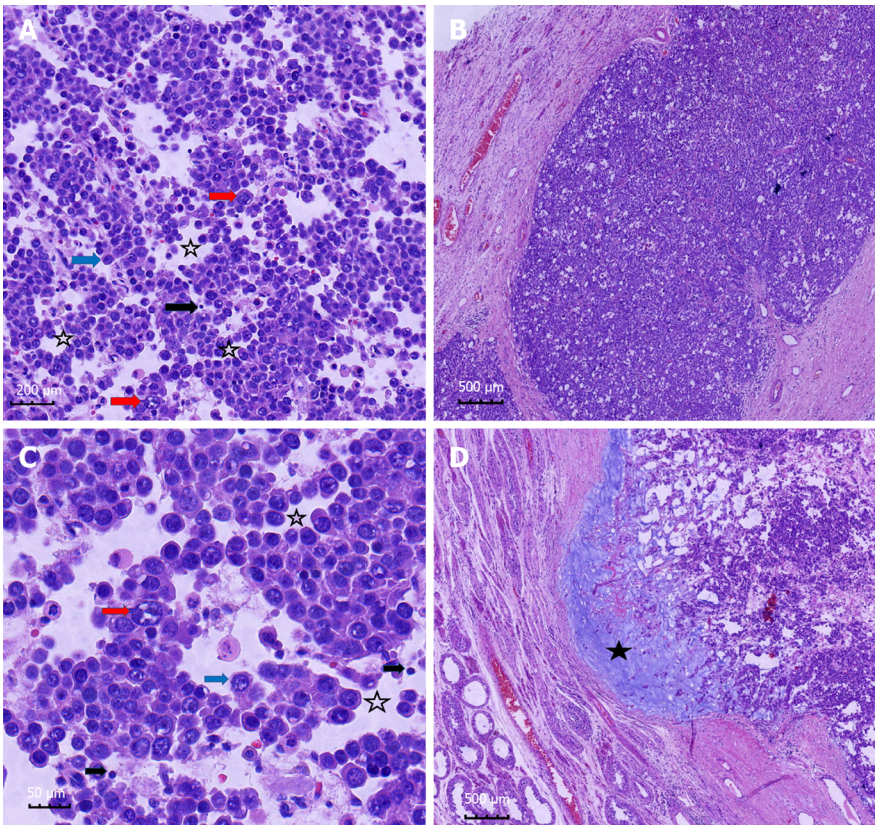
The patient agreed and voluntarily underwent orchiectomy after 1 wk. The testicular specimens were fixed with 4% formalin for 24 h. Gross examination of the right testis identified a well-demarcated, solid, lobulated, expansile, yellow-gray nodule that was 3 cm × 3 cm × 2 cm in size. The mass contained multiple areas of myxoid degeneration. The postoperative specimens were made into wax blocks and observed after hematoxylin-eosin staining. Microscopy revealed a pseudo-glandular appearance of the tumor tissue, in which aggregates of tumor cells were arranged in an edematous stroma containing scant fibrous tissue (Figure 2A and C). The tumor cells formed diffuse sheets and nests (Figure 2B). Large, small, and medium-sized tumor cells were identified. The medium-sized cell type predominated and was characterized by eosinophilic cytoplasm, suggesting decreased glycogen, round nuclei, and a filamentous chromatin pattern similar to that of spermatocytes (Figure 2A and C). The small cells resembled lymphocytes and had no obvious cytoplasm (Figure 2A and C). The large mononuclear tumor cells had round, oval, or indented nuclei, with thick chromatin and multiple nucleoli (Figure 2A and C). No mitotic figures were observed. Mucinous degeneration was observed in some areas (Figure 2D). No lymphocyte infiltration or granulomatous inflammatory response was noted. Thereafter, we performed indirect immunohistochemical staining and used mouse anti-human primary antibody and rabbit anti-mouse secondary antibody. The results revealed: CD117(+) (Figure 3A), PLAP(-) (Figure 3B), CD30(-), HCG(-), SOX-2(-), CK(-), CD45<sub>RO</sub>(-), CD3(-), D2-40(-), CK8/18(-), AFP(-), and GPC3(-). The proliferation index (Ki-67) was 80%.

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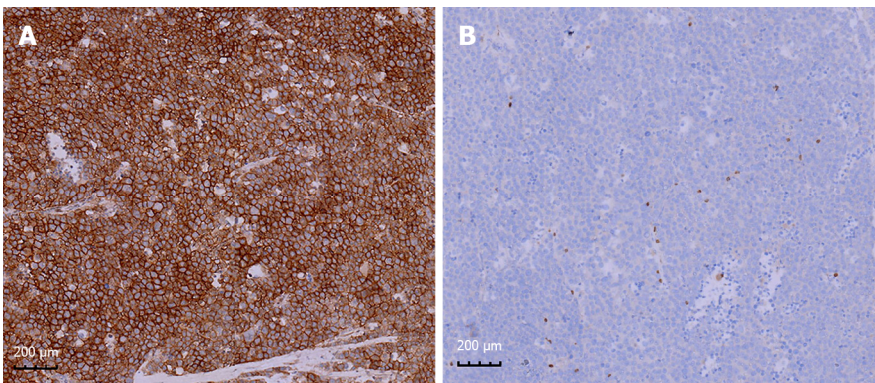
## OUTCOME AND FOLLOW-UP

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The patient only underwent orchiectomy without radiotherapy or chemotherapy. No recurrence or metastasis has been observed till date (12 mo post surgery).



**Figure 2 Histopathology findings.** A and C: Neoplastic testicular tissue with stromal edema (hollow pentagram) that produces pseudo-adenomatous change; three sizes of tumor cells were identified: Large (red arrow), small (black arrow), and medium-sized cells (blue arrow); B: Tumor cells form diffuse sheets and nests; D: Mucinous degeneration occurs in some areas (solid pentagram). Hematoxylin & eosin staining. Magnification: 100 × (A); 40 × (B and D); 400 × (C).



**Figure 3 Immunohistochemistry staining.** A: Positive staining of tumor cell membranes for CD117 (100 ×); B: No nuclear staining for PLAP (100 ×).

## DISCUSSION

The theory that spermatocytic tumor precursor cells originate in embryogenesis is disputed[3]. Some scholars[1] believe that spermatocytic tumor develops from mature cells such as pachytene spermatocytes. A recent study found that these tumor cells express reproductive cell-specific markers[4]. Morphological and immunohistochemical features of the tumor cells suggest derivation from spermatogonial stem cells [5].

Clinical symptoms associated with spermatocytic tumor include painless and slowly progressive testicular swelling[6,7] and low back pain in cases with a poor prognosis due to retroperitoneal metastasis. Spermatocytic tumors range from 2 cm to 20 cm in diameter, with an average diameter of 7 cm. Grossly, Hu *et al*[7] reported that these lobulated tumors have a homogeneous parenchymal appearance and most tumors were pink-tan, brown-tan, or white-tan and typically soft and lobulated, mucinous. They often contain areas of edema, hemorrhage, and necrosis. The majority of these

tumors are contained within the testis and do not breach the testicular sheath to infiltrate surrounding tissues[8-10]. The histomorphologic spectrum of spermatocytic tumor[11] is characterized by several points as follows: (1) At low magnification, the tumors are mainly multinodular or diffuse. All tumors have typical cell populations of three different sizes; (2) spermatocytic tumors often show edematous or myxoid degeneration with edematous stroma forming slit like structures and follicular like or irregular patterns, which are seen in some classical seminomas and rare in lymphomas; (3) tumor nodules focally show fibrous margins, and closely anastomosing connected island like structures; and (4) there is marked lymphocytic infiltration with granulomatous inflammation. It is important to recognize all the above characteristics, which will help the pathologist to diagnose and make a differential diagnosis[5]. Notably, its immunohistochemical markers are also very special. Although it belongs to germ cell tumors of the testis, it does not express useful markers in other germ cell tumors, such as OCT3/4, PLAP, AFP, HCG, and CD30. It often shows positive or weak positive expression of a key marker, CD117, and the proliferation index (Ki-67) tended to be very high. This adds challenges to our diagnostic work and requires pathologists to constantly expand their diagnostic ideas.

Spermatocytic tumor must be distinguished from classic and anaplastic seminoma. Classic seminoma is characterized by an earlier average age of onset (30 years)[2] and tumor cells that are often rich in glycogen with clear cytoplasm. The tumor cells form nests that are rimmed by fibrous tissue bands. Lymphocytic infiltration and a granulomatous inflammatory response are seen in the stroma. Classic seminoma tumor cells are positive for the immunohistochemical markers such as PLAP, CD117, vimentin, LDH, ferritin, and germ cell antigen, but usually negative for high molecular weight keratin and CD30. Anaplastic seminoma cells display obvious heteromorphism and increased mitotic rate, but do not stain positively for the abovementioned immunohistochemical markers[12]. Some experts proposed that classic seminoma and anaplastic seminoma are variants of the same tumor[13]. An elevated serum HCG level and/or tumor features, such as hemorrhage, necrosis, and vascular infiltration, are seen in patients with invasive, late clinical stage anaplastic seminoma that has a poor prognosis[14]. The spermatocytic tumor we identified in our patient contained small, medium, and large tumor cells that were slightly separated within an edematous stroma, creating a pseudo-glandular tissue appearance. The lack of lymphocytic infiltration, granulomatous inflammatory response, increased level of mitosis, and sarcomatous components were features of our patient's tumor that were consistent with this diagnosis. The tumor in our patient's case lacked markers (OCT3/4, AE1, AE3, and CD30) that are absent in spermatocytic tumor, but present in other germ cell tumors. Moreover, immunohistochemical staining for CD117 was positive in our patient's tumor; this marker is a key feature for identifying spermatocytic tumor.

The clinical features and imaging findings of testicular spermatocytic tumor are not distinct from those of classic spermatoplasm cytomaspertocytic tumor and other types of testicular tumors. Laboratory test results for serum LDH, HCG, and AFP levels are usually not elevated. Ultrasound is useful for early testicular tumor detection and is a preferred imaging method due to its non-invasive nature[14-18]. CT provides more information regarding features such as tumor boundary, internal architecture, involvement of surrounding structures, and lymph node metastasis, and is useful for establishing a preoperative presumptive diagnosis and for postoperative follow-up[19-22]. However, biopsy and histopathology are required to confirm the diagnosis. Currently, orchiectomy is the preferred treatment for testicular spermatocytic tumor. The scope of surgical resection should include the testicular epididymis and a portion of the spermatic cord to ensure complete excision for the 10%-15% of testicular tumors that invade these structures[23-26]. Most spermatocytic tumors exhibit benign behavior, with a low potential for invasion and metastasis. Given the favorable prognosis for this tumor type, no adjuvant therapy is usually required following excision by orchidectomy[27,28]. However, tumors that are larger than 4 cm in diameter, involve the epididymis or spermatic cord, or have sarcomatous features may have an increased risk for malignant behavior and recurrence. Long term follow-up is necessary in such cases.

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## CONCLUSION

In summary, we report a typical case of spermatocytic tumor, review its epidemiology and various examined features, as well as pathological features such as three typical cellular morphologies and interstitial mucinous degeneration, and make a differential

diagnosis with its similar counterpart. The first line of therapy for spermatocytic tumor is orchiectomy, which does not require other adjuvant therapy unless there is an incomplete or spermatic cord, or recurrent features. Long-term follow-up may be recommended to identify potential postoperative recurrence.

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