Radiology Case Reports 18 (2023) 3572–3576

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journal homepage: www.elsevier.com/locate/radcr

Case Report

Spermatocytic tumors in 2 patients aged 50 and 77 years: 2 case reports and brief review of the literature ☆

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ARTICLE INFO

Article history:
Received 12 July 2023
Revised 18 July 2023
Accepted 19 July 2023

Keywords:
Testicular cancer
Ultrasound
Spermatocytic tumor
Spermatocytic
Urogenital cancer
Scrotal tumor

ABSTRACT

Testicular cancer is predominantly diagnosed in young men aged 15-35 years. However, there are some rare tumors such as spermatocytic tumors that are seen more often in the older male population. Spermatocytic tumors have previously been known as spermatocytic seminomas in the scientific literature. We report the cases of 2 patients aged 50 and 77 years both diagnosed with spermatocytic tumors. In this paper we will discuss the ultrasound and histopathology features of these tumors and review the literature of spermatocytic tumor cases.

Introduction

Spermatocytic tumors are rare, typically they have a slow growth and are not associated with pain. Overall, they account for approximately 1% of all testicular tumors [1,2] with an estimated incidence of 0.2 per 100 000 [3]. Previously, the literature reported spermatocytic tumors as spermatocytic seminomas due to similar appearances, however today spermatocytic tumors are not considered a seminoma subtype. Classic seminomas are seen in younger men from 15 to 35 years old. There are fundamental differences between seminomas and spermatocytic tumors, for example, spermatocytic tumors tend to have a low growth rate [4].

☆ Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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https://doi.org/10.1016/j.radcr.2023.07.043

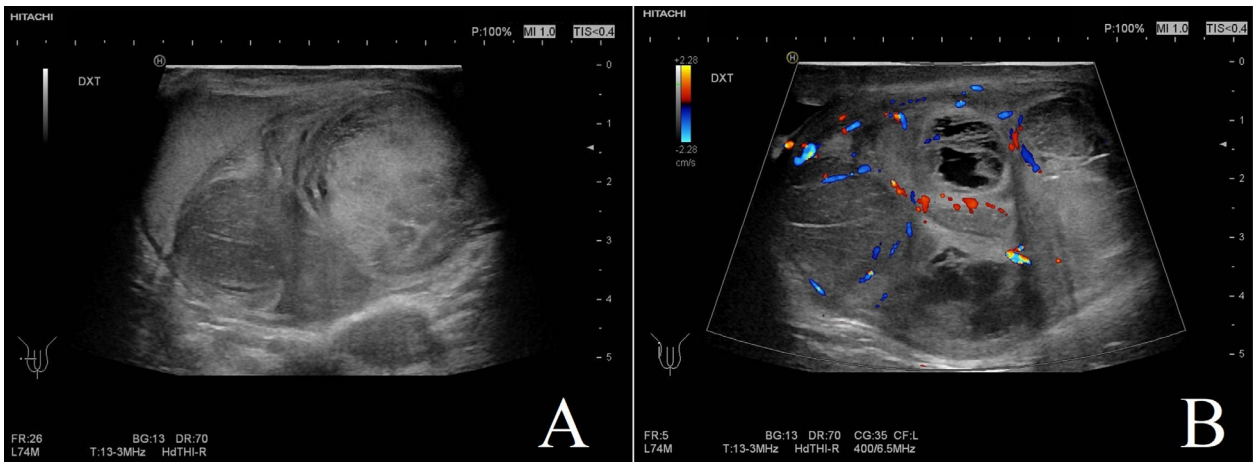

Fig. 1 – (A) Ultrasonic appearance of a right-sided tumor measuring 6 x 4 x 6, 4 cm in size in the 50-year-old patient. (B) Color Doppler of the spermatocytic tumor.

Most case reports and reviews describe spermatocytic tumors in the population older than 50 years [2,5,6], but the tumors are also seen in younger males. Cryptorchidism is associated with testicular germ cell tumors, but not spermatocytic tumors [7]. Usually, patients diagnosed with spermatocytic tumors have an excellent prognosis. Patients will be offered an orchiectomy including imaging follow-up because this type of tumor rarely metastasizes. Yet, a recent review found metastases in 7% of patients with spermatocytic tumors [1]. Currently there are no specific follow-up and diagnostic test recommendations for patients with spermatocytic tumors. A recent review suggested offering long-term follow-up for rare testicular tumors [8].

Case reports

Case 1

A 50-year-old man was referred to the department of urology on suspicion of testicular cancer. His medical history included ulcerative colitis, for which he had been treated for many years, and malignant melanoma 13 years prior with complete recovery. The patient had experienced right-sided scrotal enlargement for 2 months, and his general practitioner (GP) had palpated a large right-sided testicular mass. He had no related symptoms but reported a current exacerbation of ulcerative colitis.

At the time of referral, he had normal laboratory values, including normal tumor markers alpha-fetoprotein (AFP), $3.4*10^3$ IU/L (reference range $0\text{-}7*10^3$ IU/L), and beta-human chorionic gonadotropin (β-HCG), 0.6 IU/L (reference range 0-5 IU/L). Lactate dehydrogenase was also normal (167 U/L) (reference range 105-205 U/L). The patient was referred to a scrotal ultrasound from the urologist prior to an outpatient appointment.

Ultrasonography was performed the day after the patient's visit to the GP, and confirmed the presence of a large tumor occupying the majority of the right testicle (Fig. 1). The tumor appeared as a combination of multiple compartments, displaying both solid components with heterogeneous echogenicity and smaller anechoic cystic components. It exerted pressure on the surrounding tissues, causing a mass effect. The size of the tumor was measured to 6 × 4 × 6.4 cm. Additionally, the tumor exhibited significant Doppler signal, indicating blood flow within the mass. The left testicle appeared normal. There were no ultrasonic signs of scrotal infection.

Due to the strengthened suspicion of testicular cancer, the patient underwent right-sided orchiectomy and concurrent biopsy from the left, sonographically normal testicle. Surgery was uncomplicated and he was discharged later on the day of surgery. A CT thorax-abdomen 4 days postsurgery showed no signs of metastatic disease, but signs of pancolitis corresponding to the patient's experience of exacerbation in ulcerative colitis.

Macroscopy showed a multinodular tumor with a diameter of 59 mm and areas of bleeding and myxoid changes. There was a close relation to the hilus area as well as tunica albuginea. Microscopy revealed solid tumor areas divided by fibrous sheets, comprised by 3 tumor cell types: small cells with round, uniformly dense nuclei and scant cytoplasm; intermediate-sized cells with round nuclei and fine granular chromatin; and giant cells containing multiple nuclei with similar features to those of the intermediate cells (Fig. 2). There were no necrosis and microscopically there were no involvement of the hilus or tunica albuginea. Several mitosis and apoptotic cells were present, the ki-67 index was 50% in hotspots. The embryonic germ cell tumor immunohistochemistry markers were negative (OCT4, AFP, HCG, PLAP, CD30, and GLYP3), CD117, SALL4, and CD99 showed a strong positive reaction. The tumor was classified as a spermatocytic tumor based on immunohistochemistry as well as morphology.

Because of the radical excision of the tumor and no metastatic disease, no oncological treatment was needed. The patient has been scheduled for CT thorax-abdomen and scrotal ultrasound every 6 months for 2 years to exclude relapse of testicular disease.

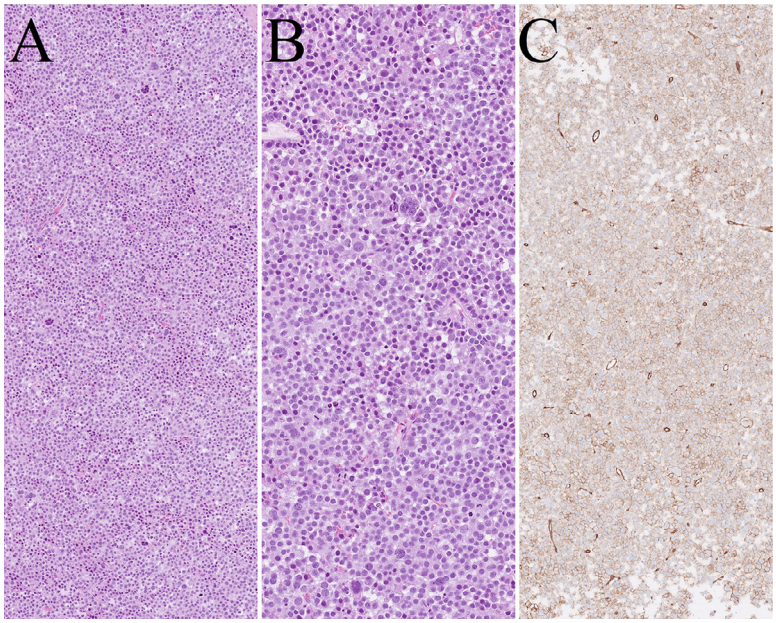

Fig. 2 – (A) H&E section of the tumor, 100× magnification. (B) 200× magnification highlighting the 3 tumor cell types. (C) Positive immunohistochemical staining for CD99.

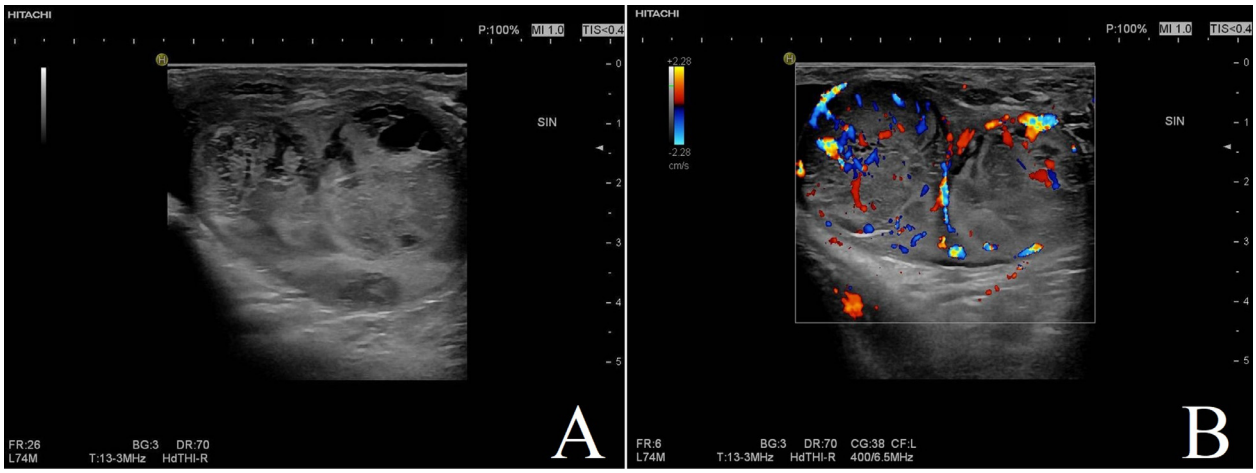

Fig. 3 – (A) Ultrasonic appearance of a left-sided tumor in the 77-year-old patient. (B) Color Doppler of the spermatocytic tumor.

Case 2

A 77-year-old man was admitted to the emergency department with a 3-day history of hematuria and fever up to 40°C. He had also experienced swelling and tenderness of the left testicle for 3 months. His medical history included hypertension, type 2 diabetes, stroke, bilateral nephrolithiasis, and previous episodes of pyelonephritis.

On admission, he had moderately elevated laboratory values for infection and impaired kidney function (eGFR 35 mL/min, reference >59). Based on the medical history and current symptoms, epididymitis and pyelonephritis were suspected, and a noncontrast abdominal CT of the kidneys and urinary tract was performed along with a scrotal ultrasound. The CT scan showed a kidney stone in the right kidney, unchanged from a CT performed a year earlier, and was otherwise unremarkable.

Scrotal ultrasound showed the left testicle to be enlarged comprising multiple tumor-like masses with heterogeneous echogenicity, possibly a single large multilocular tumor, with minimal presence of normal testicular tissue. Some of these tumors exhibited anechoic cystic components, and the tumors displayed Doppler signal, indicating blood flow inside the tumors. Additionally, “Hitachi Realtime Tissue Elastogra-

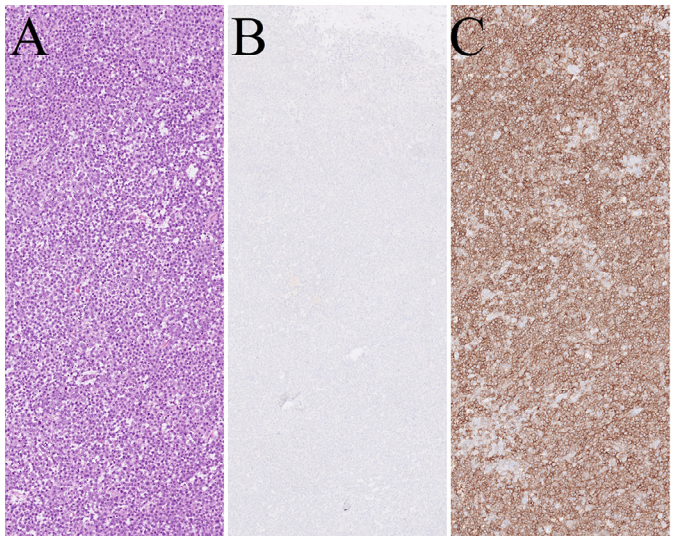

Fig. 4 – (A) H&E section of the tumor, 100 x magnification. (B) Negative immunohistochemical staining of OCT4, 100 x magnification. (C) Positive immunohistochemical staining for CD117, 100 x magnification.

phy" (HI-RTE), a type of strain elastography, areas of solid tumor showed increased stiffness compared to the surrounding tissue. The right testicle appeared normal, and no ultrasonic signs of infection were observed bilaterally (Fig. 3).

Although the referring clinician initially interpreted the radiology report as indicative of an infection, the following day it was decided to perform a left-sided orchiectomy on suspicion of testicular cancer. Surgery was performed 6 days after initial admission and was uncomplicated. On the same day, his tumor markers were normal, alpha-fetoprotein (AFP), $<2^{*}10^{3}$ IU/L (reference range 0-$7^{*}10^{3}$ IU/L), and beta-human chorionic gonadotropin (β-HCG), 0.6 IU/L (reference range 0-5 IU/L). Lactate dehydrogenase was also normal (182 U/L) (reference range 105-205 U/L). The patient was discharged the day after surgery.

Macroscopy showed a multifocal, relatively homogeneous tumor with a diameter of 31 mm, well circumscribed with a pale gray cut surface and without involvement of the hilus area or the tunica albuginea. Microscopy revealed solid tumor areas divided by fibrous sheets, comprised by 2 tumor cell types: small cells with round, uniformly dense nuclei and scant cytoplasm; and intermediate-sized cells with round nuclei and fine granular chromatin. Several mitoses were present. The embryonic germ cell tumor immunohistochemistry markers were negative (OCT4, AFP, HCG, PLAP, CD30, and GLYP3) and only CD117 and SALL4 showed a strong positive reaction. Although there were no giant cells the tumor was classified as a spermatocytic tumor based on immunohistochemistry as well as morphology (Fig. 4).

One month after the surgery, a follow-up noncontrast CT showed no signs of metastatic disease.

Because of the histopathological diagnosis and results of the CT scans, the patient has not been scheduled for follow-up scans in relation to the tumor. Contralateral testicular biopsy was not performed initially due to recent infection and has not been scheduled. A follow-up scan for the kidney stone has been scheduled 1 year after surgery.

Ultrasound equipment

In both cases, scrotal ultrasound was performed in using a "Hitachi Hi Vision Ascendus" medical ultrasound machine. The probe used was a "Hitachi Linear EUP-L74M Breast." This probe has a frequency range of 5-13 MHz intended for examining breast, thyroid, small parts, and other superficial structures.

Discussion

We reported 2 cases of spermatocytic tumors identified on ultrasound on the same day in our radiology department. Spermatocytic tumors are very rare and the origin has only relatively recently been determined [9]. It has also been debated whether the tumor is malignant or benign, but metastatic disease has been reported in extremely rare cases [10].

Both patients described in this paper experienced swelling of the affected testicle with no other symptoms, both had a radical orchiectomy performed and both patients had no signs of metastatic disease, all typical characteristics of the cases reported in the scientific literature.

Very few of the reported cases have included symptoms other than painless scrotal enlargement. These cases have involved associated testicular pain and ipsilateral varicocele [1,11].

Most reports do not describe sonographic findings in detail, but enlargement of the affected testicle with heterogeneity, multinodularity, hypervascularity, and cystic areas are frequently described [9,11–13]. Both tumors reported in this paper were heterogeneously echogenic with cystic areas and Doppler activity suggestive of hypervascularity. Xue et al. examined the value of contrast-enhanced ultrasound in 13 testicular tumors, one of which was a spermatocytic tumor [14]. All tumor types showed abundant blood flow on conventional ultrasound, which was generally unable to discriminate tu-

mor types, but with contrast-enhanced ultrasound, the spermatocytic tumor was the only type to show low enhancement. Enhancement was further characterized as uneven and fast-forward with equal regression [14]. Although contrast-enhanced ultrasound may thus assist in differentiating between tumor types, the clinical implications of this discrimination are uncertain since patients will undergo orchiectomy with subsequent immunohistochemical examination for final diagnosis regardless of sonographic tumor characteristics. Ultrasound is important in identifying the tumor and because the ultrasonic appearance of spermatocytic tumors is not different from that of other malignant testicular tumors, we assume that ultrasound has a high sensitivity for identifying spermatocytic tumors.

The 2 patients reported here had normal tumor markers AFP and bHCG and normal LDH. The same applies to almost all the cases reported in the scientific literature except one with elevated AFP, 2 with elevated bHCG, and 4 with elevated LDH [1,9]. This suggests a low tendency for the spermatocytic tumor type to exhibit elevated tumor markers otherwise often associated with testicular cancer [1,9,10,12,13,15].

As to histopathology, the 2 cases illustrate the diversity of this entity in both gross appearance and histomorphology, Case B only demonstrating 2 cell types. The 2 cases were evaluated in 2 institutes ensuring the diagnosis and despite the age variation and the different morphological appearance, the classic immunohistochemical profile supported the diagnosis [10].

Conclusion

Spermatocytic tumors are very rare, especially in men below 50 years old, and typically present with monosymptomatic swelling of the testicle. Radical orchiectomy is the treatment of choice and final diagnosis is reached by immunohistochemical examination, as sonographic and other radiologic characteristics have not been sufficiently established to differentiate between spermatocytic tumors and other testicular neoplasms. However, ultrasound can be assumed to have a high sensitivity for spermatocytic tumors as these mimic other malignant testicular cancers sonographically.

Both patients have not experienced relapse of testicular disease.

Authors' contributions

All authors contributed to this work. All authors have read and approved the final version of the manuscript.

Ethics approval and consent to participate

Our institution does not require ethical approval for reporting individual cases or case series.

Patient consent

Written informed consent for publication was obtained from the 2 patients. The authors will provide copies of these written consents upon request from the journal.

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