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MRI of a Sertoli Cell Tumor of the Testis: A Case Report

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Abstract

Introduction: Sertoli cell tumors (SCTs) of the testis are relatively rare neoplasms that originate from the sex cord and interstitial stromal cells of the testis. Magnetic resonance imaging (MRI) can be used for characterization, particularly recommended when clinical and ultrasonography (US) features are indeterminate for the staging of local lesions. Scrotal MRI may also help in the imaging-based differential diagnosis of a wide range of testicular neoplasms and prevents unessential radical orchiectomy in cases of benign lesions. Accurate and early diagnosis will play a key role in keeping the viability of testicle and fertility.
Case Presentation: We present the case of a 46-year-old male with a 1-year duration of a left painless testicular tumor that has had some recent enlargement. He has been admitted to the Department of Urological Surgery, Shenzhen Bao'an people's hospital, Shenzhen, Guangdong Province, China, on Dec 2nd, 2018. Laboratory tests were within normal limits. A well-defined margin (pseudocapsule) can be found both on US and pre-contrast MRI, but organ-sparing surgery is not advocated for the suspicious malignant features, including recent enlargement, intratumoral hemorrhageand necrosis, and contrast-enhancing solid tissue with indistinct and irregular marginss on MRI. Finally, this patient proceeded with radical orchiectomy after a sufficient preoperative assessment.
Conclusions: The low incidence rate of SCTs and their similar clinical features to those of other testicular tumors make their preoperative diagnosis challenging, but the identification of the tumor's extent, morphologic information, signal component, adjacent relationship, and distant metastasis on MRI images can be vital to make a differential diagnosis, establish reasonable intervention plans, and predict the prognosis.

Keywords: Sertoli Cell Tumor, Testis, Ultrasonography, Magnetic Resonance Imaging

1. Introduction

The main histologic categories of primary testicular neoplasms are testicular germ cell tumors (GCTs), comprising approximately 90-95% of the total tumors (1, 2). Only approximately 4% of testicular tumors originate from the sex cord-stromal cells (1, 2). Leydig cell tumors represent the majority (75%) of the common histologic types, while the remainder are much less common (1, 2). Sertoli cell tumors are relatively rare, accounting for less than 1% of all testicular tumors (3). There are three distinct subtypes of SCTs, including classical, sclerosing, and large cell calcifying (2). Although these tumors can occur in any age group, even in infants, the prevalence of SCTs tends to be higher in males aged 30 - 40 years old (3). Because of the low incidence, the etiology and biology of Sertoli cell tumors remain to be elucidated, but current clinical evidence suggests that there are many factors that increase the risk of developing these tumors (4). For example, there is a strong correlation with risk factors, including family history, cryptorchidism, and pseudo-hermaphroditism (4, 5).

The most common clinical presentation of an SCT is a painless, firm, unilateral neoplasm in an enlarged testicle, which may be found on conventional physical examination, or patients may discover it by self-examination and seek further evaluation. Usually, the patient will mention recent trauma to the scrotum or testicles; however, this trauma is not the cause of mass growth; it only makes the patient realize the testicular tumor that is already present. A few patients may present with pain within the scrotal mass, often due to infarction or hemorrhage of the tumor. Levels of serum tumor markers, including beta-human chorionic gonadotropin (β-hCG), alpha-fetoprotein (AFP), follicle-stimulating hormone (FSH), and luteinizing hormone (LH) are often negative (4). As aromatase production is the responsibility of Sertoli cells, the secretion level of estrogen may be elevated, in which case patients would present with nipple tenderness or gynecomastia.

The low incidence of SCTs and their similar clinical features to those of other testicular tumors make preoperative diagnosis challenging. However, MRI, including

a multi-parametric protocol, has emerged as a valuable technique for supplying additional information to better characterize testicular tumors. It has been recommended by the ESUR Scrotal and Penile Imaging Working Group, particularly when clinical and US features are controversial for the staging of local lesions (6). Scrotal MRI may also help in the imaging-based differential diagnosis of a wide range of testicular neoplasms and prevents unessential workup and radical orchiectomy in cases of benign lesions. Accurate and early diagnosis will play a key role in keeping the viability of testicle and fertility. To the best of our knowledge, limited MRI manifestations of SCTs have been reported in the relevant literature. Previous reports mostly have highlighted US, CT, or pathological findings in different subtypes of SCT (see Table 1 for details). Here, we present the case of a newly diagnosed man with intratesticular SCT, whose clinical and US features were nonspecific and controversial for clinical decision. We utilize this case from our institution to show the tumor's characteristic findings on MRI for helping to narrow down the differential diagnosis and to determine more precise treatment strategies.

2. Case Presentation

A 46-year-old male first presented to the Department of Urological Surgery in Shenzhen Bao'an people's hospital (The Affiliated Hospital of Shenzhen University, Shenzhen, Guangdong province, China) on Dec 2nd, 2018, with a 1-year history of a left painless testicular mass that was reported to have grown slightly during the past month. He denied any other symptoms, such as night sweats, weight loss, etc. The patient's past medical history was negative, including for cryptorchidism. The vital signs of the patient on admission were as follows: temperature, 36.6°C; heart rate, 82 beats/min; and blood pressure, 120/75 mmHg. Clinical physical examination showed a mild, painless swelling of the left scrotum with partial transillumination. He had a palpable firm mass located in the left testis. There was no palpable inguinal lymphadenopathies, skin lesions, or gynecomastia. There were no abnormal changes in the rest of the genital examination. Serum tumor makers for testis cancer, including β-hCG, AFP, and sex steroids, including FSH, LH, testosterone, estrogen, and progesterone, were drawn and found to be all within the normal ranges, and there was no sign of endocrine abnormality. Other basic laboratory studies were obtained as well and were unremarkable (e.g., complete blood count and kidney and liver function tests). The findings in the initial assessment and laboratory tests of this patient are shown in Table 2.

Table 2. The Initial Assessment and Laboratory Test Results of the Patient

Evaluation	Results
Temperature, °C	36.6
Heart rate, beats/min	82
Blood pressure, mmHg	120/75
Serum tumor makers	Normal
β-hCG, mIU/mL	< 1.2
AFP, ng/mL	2.42
Sex steroids	Normal
FSH, mIU/mL	13.68
LH, mIU/mL	5.78
testosterone, ng/mL	380.2
Estrogen, pg/mL	38
progesterone, ng/mL	0.50
Complete blood count	Normal
Kidney and liver function tests	Normal

Scrotal ultrasonography (EPIQ5, Philips) showed a well-defined, mixed solid and cystic lesion with mixed echogenicity in the swollen left testicle, measuring approximately 28 × 22 × 17 mm (Figure 1A and B). The cystic area varied from anechoic to echogenic in appearance (Figure 1B). The contralateral testis was normal. The mass revealed peripheral and minimal internal blood flow on color doppler flow imaging (CDFI) (Figures 1C and D).

A pelvic MRI was recommended for further evaluation of the testicular neoplasm. The MRI (MAGNETOM Skyra 3T MRI Scanner, Siemens) showed a complex tumor containing both solid and cystic components in the swollen left testis measuring 2.5 × 2.0 × 2.7 cm (Figure 2A-C) with a distinct margin for the existence of a pseudo-capsule, which is depicted as a hypointense halo on short-time inversion recovery (STIR) and T2WI (Figure 2A and C). The solid component showed an isointense signal on T1 weighted images with a patchy high signal (Figure 2B) and a hypointense signal on STIR, T2WI, and diffusion-weighted imaging (DWI) (Figures 2A, C and D). The front cystic area of the tumor demonstrated low T1 signal intensity and high T2 signal intensity homogeneously (Figure 2B and C). Post-contrast images also showed heterogeneous enhancement of the solid component with indistinct and irregular margins on its peripheral rear part, and the cystic region failed to demonstrate enhancement (Figure 2E).

After a preoperative evaluation based on clinical and imaging examinations, the patient proceeded with an radical inguinal orchiectomy. The gross specimen included the testicle with the attached spermatic cord measuring 6 × 3 × 2 cm. One mixed cystic and solid nodule with necrosis and a gray to red cut surface measuring 2 × 1 × 1 cm was identified in the testicle. Histologic examination revealed

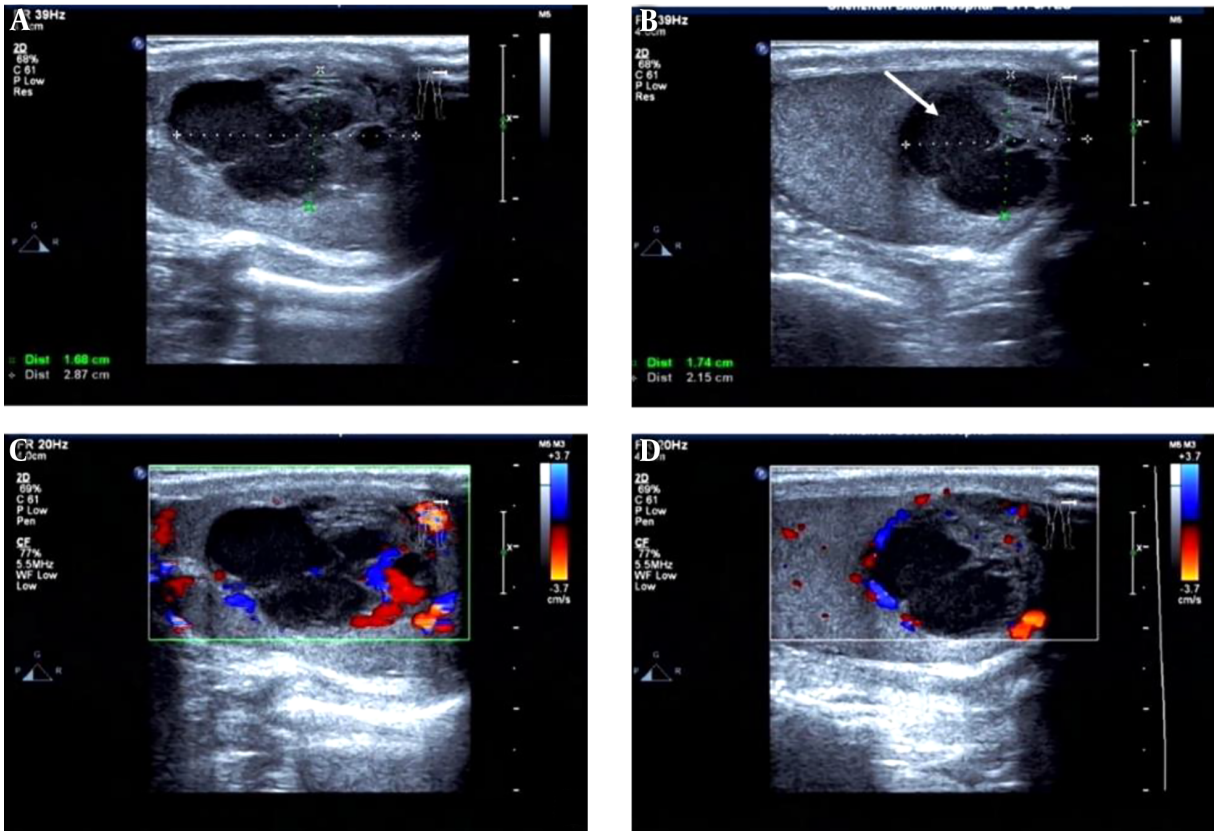

Figure 1. Scrotal ultrasound and color doppler flow images. Scrotal ultrasound showed a well-defined, mixed solid, and cystic lesion with mixed echogenicity in the swollen left testicle (A); the cystic area varied from anechoic to echogenic in appearance (B white arrow). The mass revealed peripheral and minimal internal blood flow on color doppler flow imaging (C and D).

a hollow tubular arrangement of uniform tumor cells in a round or oval shape with ample eosinophilic cytoplasm, no prominent nucleoli, and rare mitotic figures (Figure 3A). Dilatation of the blood sinus was observed in the interstitial stroma (Figure 3B). The tumor was positive for cytokeratin (CK) (Figure 3C), P53, CD99, S-100 (Figure 3D), and vimentin (Figure 3E), while it was negative for AFP, CD117 (c-kit), HCG, OCT3/4, PLAP, calretinin (CR), inhibin-α, and SMA on immunohistochemical staining. The label index was approximately 5% positive for Ki-67. The morphologic and immunohistochemical findings suggested the diagnosis of a classical Sertoli cell tumor (ICD-O code: 8640). At the time of the 6-month follow-up visit, the scrotal ultrasounds showed no evidence of recurrent or residual tumor.

3. Discussion

SCTs are generally considered to be benign, with the majority of cases confined to the testicle parenchyma at diagnosis. However, as the current largest cohort study data suggested, 35% of SCT patients are thought to have metastatic potential, and SCTs follow a more aggressive malignant course compared to that of LCTs (3).

Scrotal ultrasonography is the initial preferred imaging modality for a suspected testicular mass (4). The primary aim is tumor localization and preliminary lesion characterization (solid, cystic or mixed). In our case, the testicular tumor was reported to have had some recent enlargement, while US findings only showed a well-defined mixed echotexture with signs of blood flow on color doppler imaging, which are indeterminate for a differential diagnosis and local staging.

MRI has emerged as a valuable second-line imaging technique for supplying additional information to better characterize testicular tumors, particularly recommended when clinical and ultrasound features are controversial for the staging of local lesions (1, 6). Lack of ionizing radiation, less dependence on operator technique, multiplanar imaging capability, and superior soft-tissue contrast

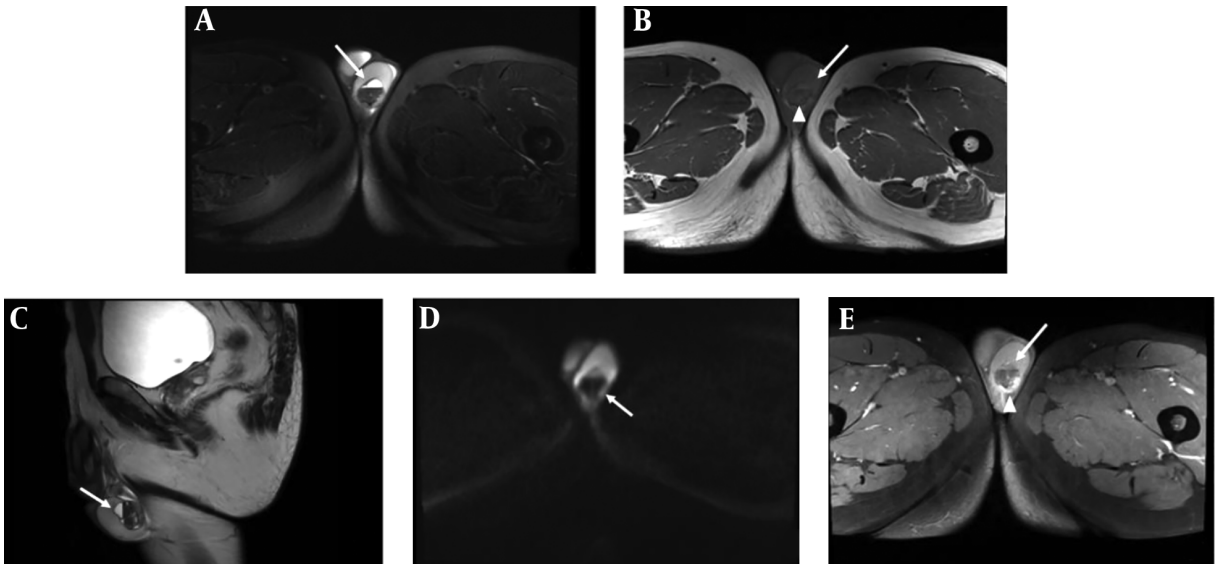

Figure 2. Pelvic Magnetic resonance images. Pelvic MRI showed a complex tumor containing both solid and cystic components in the swollen left testis with a distinct margin for the existence of a pseudo-capsule (arrows, A-C), which is depicted as a hypointense halo on STIR (arrow, A) and T2WI (arrow, C). The solid component showed an isointense signal on T1WI (arrow, B) with a patchy high signal (arrowhead, B) and a hypointense signal on STIR (A), T2WI (C) and DWI (arrow, D). The front cystic area of the tumor demonstrated low T1 signal intensity (B) and high T2 signal intensity homogeneously (C). Post-contrast images (E) also showed heterogeneous enhancement of the solid component with the indistinct and irregular margin at its peripheral rear part (arrowhead), and the cystic region failed to demonstrate enhancement (arrow).

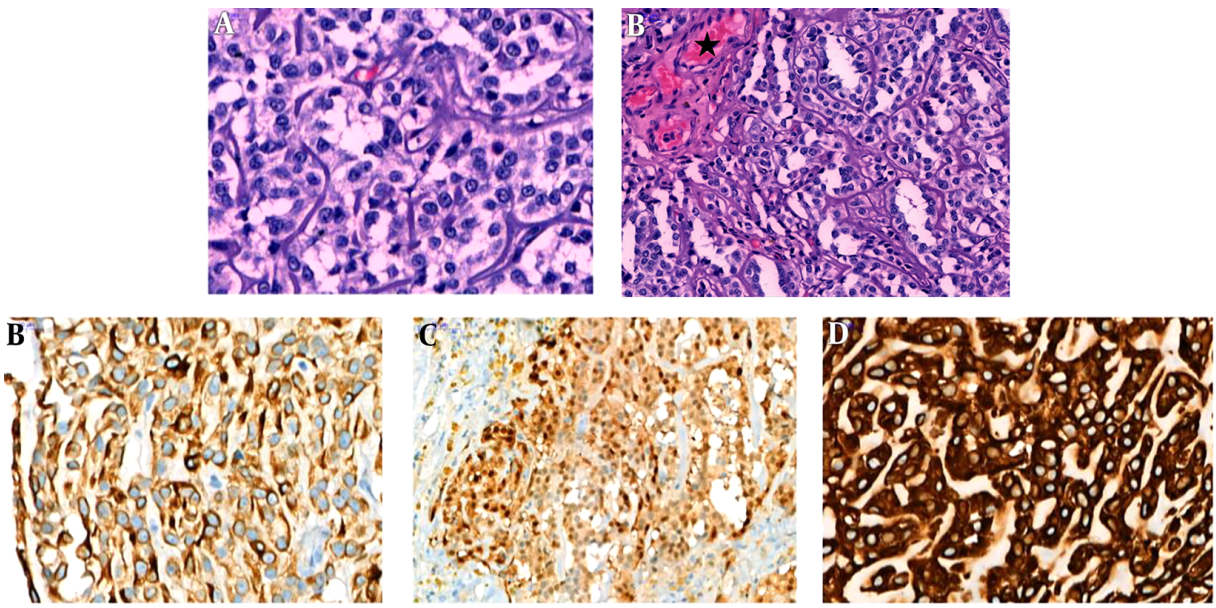

Figure 3. Pathological findings. A, Histologic examination revealed a hollow tubular arrangement of uniform tumor cells in a round or oval shape with ample eosinophilic cytoplasm, no prominent nucleoli and rare mitotic figures (hematoxylin and eosin stain, 400×); B, dilatation of the blood sinus (black star) was observed in the interstitial stroma (hematoxylin and eosin stain, 400×); C, immunohistochemistry for CK stains tumor cells; D, immunohistochemistry for S-100 stains tumor cells; E, immunohistochemistry for vimentin stains tumor cells.

resolution make MRI a perfect choice of sufficient analysis about image findings before surgery and prevent unessential invasive procedures, such as needle biopsy.

Combining the literature review with our case (see Table 1 for details), Sertoli cell tumors tend to appear as a well-circumscribed isolated neoplasm with an isoin-

tense signal on T1-weighted imaging (T1WI) and a markedly hypointense signal on T2-weighted imaging (T2WI) compared to imaging in the normal parenchyma of the contralateral testis; SCTs are mainly heterogeneous with cystic and solid components both on unenhanced and contrast-enhanced MRI. MRI findings have been proved to be closely correlated with the histopathologic characteristics of SCTs (1, 5, 9). In our case, the tumor showed a high signal on T1WI in keeping with the presence of a focal hemorrhage on pathology. Post-contrast images also demonstrated heterogeneous enhancement of the complex mass with a component of cystic and necrotic tissue. The presence of a pseudo-capsule is a characteristic marker of stromal tumors on MRI and is detected as a halo of low signal intensity surrounding the mass on T2WI. When testis-sparing surgery is planned, the pseudo-capsule is another valuable MRI marker that ensures that enucleation of the testicular mass can be performed in benign tumors in order to keep the viability of the testicle (1). However, conventional MRI features that are suspicious for malignancies of the testis include a large tumor size > 5 cm, rapid enlargement, widespread hemorrhage and necrosis, infiltrative tumor margins, invasion to the testicular tunicae and adjacent tissue, lymphovascular invasion, and distant metastasis (1). In the specific case of our patient, even if a well-defined pseudo-capsule can be found both on US and pre-contrast MRI, organ-sparing surgery is not advocated in the presence of suspicious malignant features including recent enlargement, presence of intratumoral hemorrhage (with high T1 signal) and necrosis (with high T2 signal and the lack of contrast enhancement), and solid contrast-enhancing tissue with indistinct and irregular margins on MRI. More aggressive management approaches should be proposed for patients with these high-risk features (3). Finally, this patient proceeded with radical orchiectomy after a sufficient preoperative assessment.

There is also much debate regarding the prior management of primary testicular neoplasms after partial or radical orchiectomy, especially when they follow a benign course (10). For patients with clinical stage I disease after partial orchiectomy alone, the cure rates are still very high without adjuvant therapies or retroperitoneal lymph node dissection (4). MRI findings can be helpful in reducing healthcare costs and improve patient management for these cases with a conservative approach.

MRI may also help to distinguish SCTs from other common tumors of the testicle. Compared to SCTs, GCTs usually present at younger ages (3). Typical seminomas of the testicle have been reported to be neoplasms with ill-defined margins; seminomas are mainly homogeneous, weakly hypointense on T2WI, and weakly hyperintense on T1WI. The specific feature for seminomas is the presence of fibrovascular septa on pathology, detected as hypointense band-like structures within the tumor on T2WI (1). Dynamic contrast-enhanced MRI data can be used for differential diagnosis. Usually, Sertoli cell tumors enhance heterogeneously with early and avid enhancement, followed by a prolonged washout, while seminomas enhance relatively homogeneously with weak, progressive enhancement and no washout (1). Moreover, the fibrovascular structures are found to enhance more than the rest of seminomas (1). Teratomas of the testis appear as a type of tumor with multiple signal intensities corresponding to the components of hair, calcification, and fat. Lymphoma mainly in elderly patients with systemic symptoms and nearly always shows restricted diffusion and a reduced apparent diffusion coefficient (ADC) due to increased cellularity of the tumor.

For other non-seminomatous germ cell tumors, MRI features are often not specific (1), and serum tumor markers are a useful next step for identifying tumors of testis in this category (4). In general, seminomas never have an elevation of AFP, and only approximately one-fifth of them may have an elevation of β-hCG. AFP is rarely elevated, but β-hCG will usually be elevated in choriocarcinoma. Teratomas rarely have elevations of either AFP or β-hCG. In addition, the serum levels of AFP and β-hCG may be increased in mixed germ cell tumors according to the histologic subtypes.

In conclusion, Sertoli cell tumors are a subtype of sex cord-stromal tumors of the testicle that occur relatively rarely in adult males. Based on these clinical and imaging features, SCTs should be taken into consideration in the differential diagnosis of this case. Current literature shows a risk-stratified approach to surgical treatment in patients with high-risk SCT can be helpful and may have positive implications on prognosis (3, 10). Making a precise diagnosis before surgery is challenging, but the identification of tumor's extent, morphologic information, signal component, adjacent relationship, and distant metastasis on MRI images can be vital to reach a differential diagnosis, establish reasonable intervention plans, and predict prognosis. However, long scan time is the major weakness of MRI, especially for elderly and pediatric populations, and evaluation can be harder due to patient motion. Given the limitation of our case, further studies with more data collection combining morphological and functional MR techniques will add more meaningful information to improve our findings.

Footnotes

Authors' Contribution: Yu Hu reviewed the literature and drafted the manuscript. Jian Xun Song supervised and reviewed the manuscript. Yi Chao Zhang collected the clinical data and proofread the manuscript. Xiao Ming Li were responsible for the pathological study.

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Table 1. Main Features of Our Case and Previously Reported Cases in Recent Years

Our Case or Reference	Location of lesion	Size	Clinical History	Imaging Features	Treatment	Histological Subtype of SCT or Mixed
Our case	In the left testis	2.5 cm in diameter	A 46-year-old male with a 1-year duration of a left painless testicular tumor which has had recent enlargement	A well-defined isolated neoplasm with pseudo-capsule can be found both on the US and pre-contrast MRI. MRI showed a complex tumor containing solid and cystic components. The solid component showed an isointense signal on T1 with a patchy high signal in keeping with a focal hemorrhage. Post-contrast images also showed heterogeneous enhancement of the solid component with the indistinct and irregular margin at its peripheral rear part. The region of cystic and necrotic tissue failed to demonstrate enhancement	An inguinal approach left radical orchiectomy	Classical
Bardisi et al. (5)	Right testicular lesion	12 mm in diameter	A 28-year-old male with left testis pain.	US evaluation revealed grade 3 left varicocele and an incidental 9 mm calcified mass in the right testicle, which was further confirmed by MRI	A right partial orchiectomy.	Benign large cell calcifying
Li et al. (7)	In the left testis	1.4 cm in diameter	A 7-year-old healthy boy with a 3-day history of constant left testicular pain.	US revealed a left-sided, well-defined, and heterogeneous intratesticular mass. Small echogenicities were noticed within the lesion, likely representing punctate calcifications. A shell of normal left testicular parenchyma was seen peripheral to the mass.	A left partial orchiectomy was planed, but actually radical orchiectomy	Malignant large cell calcifying
Tracy et al. (8)	The upper pole of the left testis	1.5 cm in diameter	A 13-year-old pubertal male with a three-week history of left scrotal swelling	US revealed a heterogenous encapsulated calcified lesion in the upper third of the left testis	An inguinal approach left partial orchiectomy	Benign large cell calcifying
Tahaineh et al. (9)	Left scrotal mass	11 cm in diameter	A 27-year-old man with a huge left scrotal mass of 6-years duration.	US showed large mixed solid and multilocular cystic mass in the left testis. MRI showed a large complex cystic lesion with low signal on T1WI and high signal on T2WI with a part of high signal on both T1 and T2 in keeping with hemorrhage. Post-contrast images also showed heterogeneous enhancement. CT showed no evidence of extra-testicular metastasis	An inguinal and scrotal approach left radical orchiectomy	Mixed Sertoli-leydig-granulosa sex cord tumor of testis