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Case Report

Ultrasonography of a giant adenomatoid tumor of the tunica vaginalis: A case report and review of the literature [☆]

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ABSTRACT

Adenomatoid tumor is a kind of benign tumor, accounts for a low percentage of scrotal tumors. It usually locates in epididymis, less commonly, arises from the tunica albuginea, spermatic cord or tunica vaginalis of male's urogenital tract system. Here, we report a case of a 32-year-old male, which presented as a 6-month history of left scrotal swelling pain with a scrotal mass, and it had progressively enlarged over the last 1 month. The ultrasonography (US) described a giant well-defined hypoechoic mass in the left scrotum, a thick pedicle connected the mass and the left scrotal wall, and the pedicle had large blood vessels in it. Enhanced CT showed an irregular solid mass in the left scrotum, the left testicular artery was thickened to supply blood for the mass, and the radiologist mentioned it could not be excluded as a malignant lesion. The patient underwent left radical orchiectomy, the mass was diagnosed as an adenomatoid tumor of the tunica vaginalis by pathology. We review literature regarding adenomatoid tumors originating in the tunica vaginalis, summarize the ultrasonographic presentations, provide the idea of diagnosis and differential diagnosis to improve diagnostic accuracy and avoid unnecessary orchiectomy.

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Introduction

The name adenomatoid tumor was introduced by Golden and Ash in 1945. It is a rare benign tumor that accounts for 30% of all paratesticular masses [1]. Adenomatoid tumors can occur in the urogenital tract systems in both male and female. In male, it usually locates in epididymis, less commonly, arises

from the tunica albuginea, spermatic cord or tunica vaginalis [2]. Adenomatoid tumors of the tunica vaginalis are quite rare and do not have typical imaging features. Their clinical symptoms and imaging presentations may resemble malignant tumors, so clinicians may mistake them for malignant tumors, resulting in overtreatment. As a sort of benign tumor, adenomatoid tumor of the tunica vaginalis usually undergoes local mass resection with preservation of testicular, rather than radical orchiectomy. Here, we report a case of giant adenomatoid tumor of the tunica vaginalis. We review

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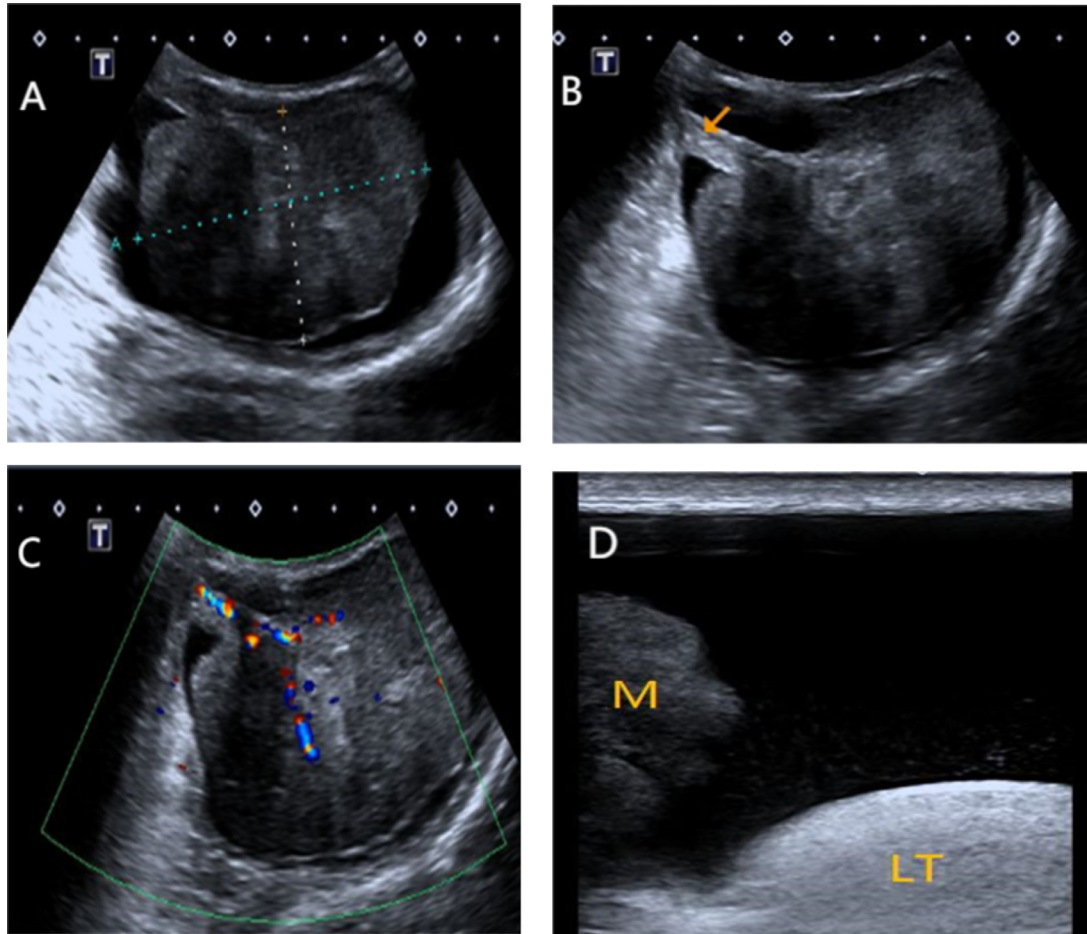


Fig. 1 – Scrotal ultrasound images. (A) A hypoechoic mass in the left scrotum measuring approximately 7.7 × 6.7 × 4.8 cm. (B) The mass connected to the scrotal wall by a thick pedicle (as the arrow shows). (C) Blood flow entered into the mass through the pedicle. (D) The left testicle was compressed.

literature to summarize the ultrasonographic presentations, provide ideas for diagnosis and differential diagnosis, and provide some insight for clinicians to help plan the best surgical approach.

Case presentation

A 32-year-old male with a 6-month history of a progressively larger left scrotal mass presented to the hospital with a painful swelling of the left scrotum. He denied having any history of infection, trauma or other tumors. On physical examination, the left scrotum was significantly enlarged and a firm mass of approximately 8.0 cm in diameter was found in the scrotum, while the penis and right scrotum were normal. Preoperative tumor markers and hormone levels, such as CEA, AFP, PSA and β -HCG were within normal ranges.

Scrotal ultrasound (US) was performed and revealed a hypoechoic mass in the left scrotum with the size of 7.7 × 6.7 × 4.8 cm (Fig. 1). In addition to this solid hypoechoic lesion, hydrocele was found in the left scrotum, the left testicle was compressed, and the left epididymis was not sat-

isfactorily displayed. The mass was irregularly shaped with well-defined borders. Notably, the mass was attached to the left scrotal wall by a thick pedicle, and blood vessels were visible within this pedicle. Color Doppler showed small amount of blood signal within the mass and abundant arterial signals within the pedicle. Ultrasonography revealed that the mass was suspected to be a tumor, but the pathologic nature needed to be further determined.

Further evaluation using computed tomography (CT) imaging, revealed an irregular solid mass in the left scrotum, the left testicular artery had thickened and extended into the mass to supply blood, and continuous significant enhancement was noted, so the possibility of a malignant lesion could not be excluded (Fig. 2).

The mass grew progressively larger over a month and could not be excluded out as a malignant lesion by CT. The patient had no fertility requirements, so he opted radical orchiectomy on the left side. During the procedure of operation, clinicians found the actual size of the mass was approximately 9.0 cm, with complete capsule and no adhesions between the mass and the left testis or epididymis, which was attached to the tunica vaginalis with a thick pedicle. Tissues of the left epididymis (5 × 2 × 1.2 cm), testis tissue (3.6 × 2.0 × 1.8 cm), part of

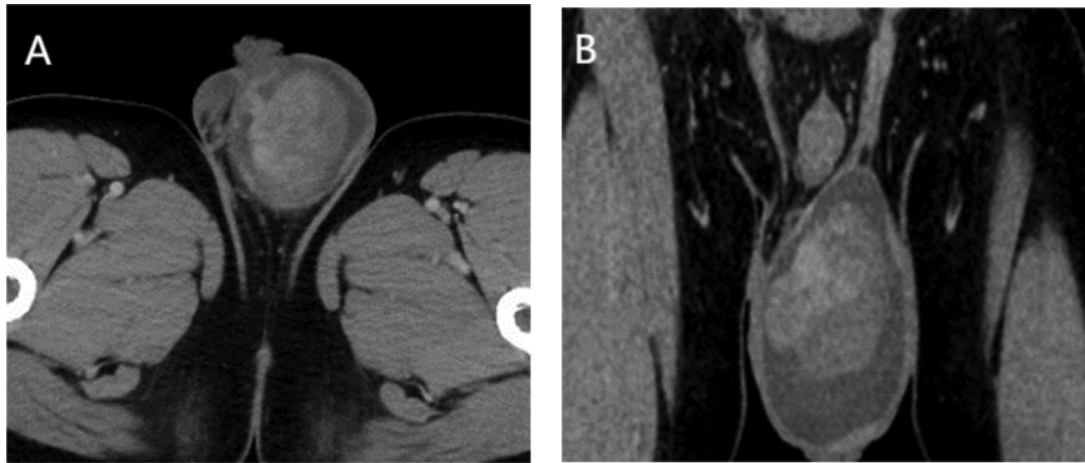


Fig. 2 – (A) Enhanced CT scan, (B) multiplanar reformation (MPR) of the mass, showing an irregular solid mass in the left scrotum, the left testicular artery had thickened and extended into the mass to supply blood.

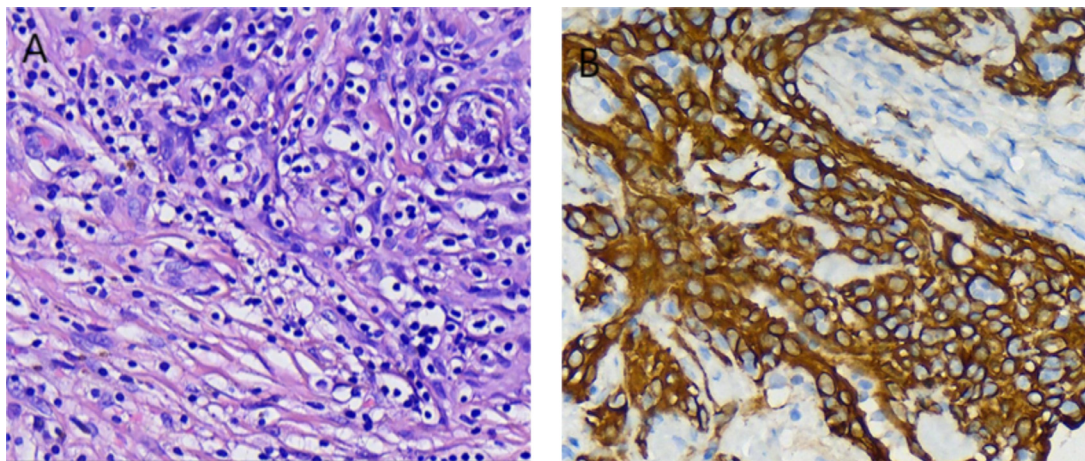


Fig. 3 – (A) The H&E staining revealed diffuse sheets of epithelioid cell and desmo-stroma structures (×40). (B) Immunohistochemistry revealed strong CK expression in neoplastic cell (×40).

spermatic cord (3.5 × 0.6 cm), and the mass (9.0 × 6.0 × 1.8 cm) were cut off for biopsy. Pathologic examination demonstrated that the lesion was composed of gland lined by flattened epithelioid cell and desmo-stroma structures. No tumor cells invaded into the epididymis, testis or spermatic cord. Immunohistochemistry showed that CK (+), CD34 (+), CR (+), EMA (+), WT-1 (+), D2-40 (+), Ki-67 (5% +) in tumor cells (Fig. 3). Therefore, histological and immunochemistry study supported the diagnosis of adenomatoid tumor of the tunica vaginalis. The clinical follow-up was performed at 3, 6, 12 and 18 months postoperatively. No recurrence of the tumor was observed.

Discussion

In male, adenomatoid tumor is a rare benign tumor usually found in the particular structures, it rarely occurs in the tunica vaginalis [3]. It is well known that scrotal adenomatoid tumors often occur in patients aged 30-50 years, and their

size usually ranges from 0.5 to 5.0 cm, and rarely exceed 5.0 cm [4], but in our case, the tumor reached over 8.0 cm, and it was an adenomatoid tumors of the tunica vaginalis, so it was very rare indeed. US is an economic noninvasive examination that is of great value in the diagnosis and differential diagnosis of scrotal tumors, and can be combined with CT scan or magnetic resonance imaging (MRI) to make a correct diagnosis. We report the case here to raise awareness of the disease.

In order to accurately diagnose adenomatoid tumors of the tunica vaginalis, we should be familiar with the anatomy of the scrotum. The testes are surrounded by the tunica albuginea and tunica vaginalis. A lesion found around testes may arise from testes, testicular appendage or the sheath. The tunica vaginalis is the continuation of the peritoneum and is divided into a visceral layer and a parietal layer. The visceral layer covers the surface of the testis and epididymis, and the parietal layer is attached to the scrotum, in a manner similar to that of the visceral and parietal pleura of the thorax. The potential space between the visceral and parietal lay-

ers, known as the testicular sheath cavity, usually contains 2-3 mL of fluid [5].

It is important to determine whether the scrotal mass is intratesticular or extratesticular, because malignant masses outside the testes are relatively rare, whereas malignant solid masses inside the testes are more common, and seminoma is the most common malignant testicular tumor in adults [6]. Although most extratesticular tumors are benign, the detection and differentiation of extratesticular tumors is actually more complex than that of intratesticular tumors. Some malignant tumors, such as malignant mesotheliomas or sarcomas may occur in the tunica vaginalis. This paper summarizes those tumors that need to be differentiated from adenomatoid tumors of the tunica vaginalis.

Malignant mesotheliomas

Malignant mesothelioma first needs to be differentiated from the adenomatoid tumor of the tunica vaginalis in our case. Most reports suggest that adenomatoid tumors originate from mesothelial cells [7], and mesothelial cells of the tunica vaginalis cause adenomas, mesotheliomas, or benign intraepithelial cysts [3], adenomatoid tumors can be considered as benign mesotheliomas. Most malignant mesotheliomas occur in the peritoneum or pleura, rarely originate from the tunica vaginalis, accounting for only 0.3%-5% of all malignant mesotheliomas [8]. Malignant mesothelioma should be suspected if the patient has a progressively enlarging mass or hydrocele, either simple or complex, and rapidly reaccumulates fluid after aspiration. In this case, the patient had a big mass with hydrocele, developed rapidly in a short period of time, these symptoms resembled a malignant mesothelioma of the tunica vaginalis.

Most malignant mesotheliomas in men occur between the ages of 55 and 75 years [9]; however, adenomatoid tumors of the male genital system often occur between the ages of 30 and 50 years [10]. Currently, the only proven risk factor for malignant mesotheliomas is exposure to asbestos, but this risk factor was only confirmed in about 35% of testicular mesothelioma cases [11]. This patient was relatively young and has no history of asbestos exposure. Ultrasonography of malignant mesotheliomas shows a simple or complex hydrocele with soft-tissue of mixed echogenic soft tissue and multiple extratesticular nodules arising from the tunica vaginalis [12], it is often accompanied by thickened tunica vaginalis, and near spermatic cord, testicular or epididymis may be invaded [8]. On color Doppler US, congestion of the involved tunica vaginalis can be seen. And enlarged lymph nodes in the inguinal area may be seen. The ultrasound findings in this case do have some similarities to malignant mesothelioma of the tunica vaginalis. However, it has no characteristics of a malignant tumor, such as a typical tunica vaginalis thickening or invasion of adjacent tissues as in malignant mesothelioma. Its borders were well-defined and there was no evidence of infiltration of surrounding soft tissues. Therefore, the diagnosis of malignant mesothelioma can be ruled out. Of course, the most important thing is to differentiate it by pathological diagnosis, and cytologic atypia can be seen in malignant mesothelioma [13].

Sarcomas of the scrotum

Sarcoma is an uncommon malignancy originating from mesenchymal cells entrapped in the spermatic cord, epididymis, or tunics. Rhabdomyosarcoma is the most common type in pediatric patients, while liposarcoma and leiomyosarcoma are the most common sarcomas in adults [14]. Ultrasonography can visualize a hydrocele and see a heterogeneous and multivascular mass that often invades the scrotum and extends into the groin area [15]. Lymph nodes involvement can be seen in one-third of paratesticular sarcomas [3].

Metastatic carcinomas of the scrotum

Metastatic carcinomas should also be distinguished from adenomatoid tumors of the tunica vaginalis, which may have similar symptoms and ultrasonographic presentations. Adenocarcinomas from the prostate, lung or gastrointestinal tract may rarely metastasize into the tunica vaginalis [16]. In fact, adenomatoid tumors of the tunica vaginalis occur in young adults, while metastatic carcinomas tend to occur in older adults. The absence of other tumors in other organs help to support that the tumor of tunica vaginalis is primary.

Tuberculosis of the scrotum

Adenomatoid tumors of the tunica vaginalis may also be confused with tuberculosis of the scrotum. The genitourinary tract is the most commonly affected site of extrapulmonary tuberculosis, scrotal tuberculosis may also present as a painless scrotal mass [17]. The US of scrotal tuberculosis usually shows local thickening of the tunics, irregular mass with mixed echo inside, but without clear borders, internal flow signals may be detected in some lesions. If the epididymis is infiltrated, the epididymal boundary is unclear on ultrasound, the internal echo is uniform, there may be hypoecho nodules in the epididymis, and no obvious blood signals is seen on color Doppler ultrasound. Irregular hyperecho and acoustic shadow can be seen in some lesions, which maybe typical manifestations on US. Some patients may have scrotal wall ulcers or sinus formation [17]. Moreover, most of these patients have renal tuberculosis and require imaging of the urinary tract.

Scrotal hemangioma

Scrotal hemangioma is very rare. Ultrasound of scrotal hemangioma shows that local thickening of the scrotal wall, with uneven internal echoes that are reticular deformed under pressure, and punctate blood flow signals can be detected on color Doppler ultrasound.

Lymphangiomas of the scrotum

Lymphangiomas are benign lesions mostly encountered at birth, 80%-90% are discovered before the age of 2 years [18]. Lymphangiomas in the scrotum is extremely rare. Scrotal masses and hydrocele are common clinical findings, similar to our case. Scrotal US showed polycystic and extratesticular mass with mixed echoes, attributable to lymphoid tissue malformation or thrombosis. Color Doppler US shows a small amount of blood flow in the septa [19].

Lipoma and leiomyoma of the scrotum

Lipomas and leiomyomas of the scrotum are rare, need to be differentiated from adenomatoid tumors of the tunica vaginalis. On Ultrasound, lipomas tend to be well-defined, homogeneous, and hyperechoic, but a hypoechoic or heterogeneous echotexture may be seen in the presence of fibrous, myxoid, or vascular tissue. There is generally no blood flow signal in the mass. Leiomyomas and adenomatoid tumors of the tunica vaginalis are very similar on US, they are both encapsulated, but a swirling pattern of echo maybe a characteristic of leiomyoma and may help differentiate it from other scrotal tumors [20].

Other paratesticular tumors

Other paratesticular tumors should be differentiated from adenomatoid tumors of the tunica vaginalis, such as the most common paratesticular adenomatoid tumors, adenomatoid tumors of epididymal. US shows a small, usually oval nodule in the head or tail of the epididymis. The most important distinguishing point is the location of these tumors.

Testicular tumor

Occasionally, when testicular tumors infiltrate the scrotal wall, such as seminomas, adenomatoid tumors of the tunica vaginalis should also be differentiated from testicular tumors. When the lesion is small, US can easily distinguish the lesion originating from the testicular or the scrotal wall according to the location of the lesion. When the lesion is large, US is very difficult to distinguish its source. So, it is important to determine whether the mass is inside or outside the testis. If necessary, ultrasound-guided needle biopsy can be performed to determine the origin.

Conclusion

In short, adenomatoid tumors of the tunica vaginalis are rare, without typical clinical features and image findings, and

should be differentiated from other scrotal tumors. The sonographic features of adenomatoid tumors of the tunica vaginalis are variable, if a pedicle is found between the mass and the scrotal wall, the possibility of adenomatoid tumors of the tunica vaginalis should be considered. In this case, if we had a correct tendentious predisposition diagnosis at the beginning, the patient would have only the mass removed, thus preserving his left testicle and appendage.

Patient consent

Patient consent was obtained for the article.

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