Journal of
Medical Case Reports

CASE REPORT

Open Access

Incidental finding of probable epididymal adenomatoid tumor and tubulocystic renal cell carcinoma in a patient with epididymo-orchitis: a case report

Alexandra Ghadieh[1], Khaled El Khatib[1], Ghassan Hamadeh[1] and Bassem Saab[1,2*]

Abstract

Background Tubulocystic renal cell carcinoma is a rare type of renal cell carcinoma (< 1%). Tubulocystic renal cell carcinoma was first acknowledged by the World Health Organization in 2016. Tubulocystic renal cell carcinoma has low aggressivity and has a metastasis rate of around 6%.

Adenomatoid tumor of the epididymis is an asymptomatic rare, benign mesothelial neoplasm of the paratesticular region, most commonly occurring at the tail of the epididymis. This tumor is prevalent in the third to fifth decades of life with no malignant potential. There have been no reported cases of both diagnoses existing at the same time in a patient.

Case presentation We aim to present the first case in literature, according to our knowledge, of both tubulocystic renal cell carcinoma and probable adenomatoid tumor existing at the same time in a 67-year-old Lebanese male presenting with epididymo-orchitis, who on ultrasound was found to have an incidental finding of probable adenomatoid tumor located in the tail of the right epididymis, as suggested by the radiologist and the urology team. The patient was also found to have a renal mass that was described earlier to be a Bosniak type 2 renal cyst. The patient underwent left partial nephrectomy. A pathological examination revealed tubulocystic renal cell carcinoma.

Conclusions Tubulocystic renal cell carcinoma might be the result of progression from Bosniak type 2 renal cysts. They are best treated by a nephron-sparing surgical approach. Physical examination and testicular ultrasound are important tools in the diagnosis of scrotal and epididymal tumors. The benignity of adenomatoid tumor of the epididymis is reassuring and is a reason why it may usually be underreported.

Keywords Renal cyst, Urological cancer, Tubulocystic renal cell carcinoma, Adenomatoid tumor

Background

The simultaneous coexistence of tubulocystic renal cell carcinoma (TRCC) and probable adenomatoid tumor is a rare occurrence that was not reported earlier in literature as per our review. Renal cell carcinoma (RCC) originates in the renal cortex and makes up 85% of all primary renal tumors. RCCs are classified on the basis of their histopathological features. TRCC is a rare type of RCC (< 1%) that was first acknowledged by the World Health Organization in 2016. They are less aggressive and have a

*Correspondence:
Bassem Saab
brsaab@aub.edu.lb
[1] Department of Family Medicine, American University of Beirut Medical Center, Beirut, Lebanon
[2] Department of Family Medicine, American University of Beirut Faculty of Medicine, P. O. Box 11-0236, Beirut, Lebanon

metastasis rate of around 6% [1]. Ultrasound, computed tomography (CT), and magnetic resonance imaging (MRI) are the principle diagnostic modalities [2]. Partial nephrectomy, excision, and ablation are the mainstay of management [2].

Adenomatoid tumor of the epididymis is a rare, benign neoplasm of mesothelial origin with no known malignant outcome [3]. It is the most common epididymal tumor (60% of benign tumors) and accounts for approximately 30% of all neoplasms of testicular adnexa, second only to lipoma [4]. Adenomatoid tumors are commonly incidental findings, most often diagnosed in patients in the third through fifth decades of life with very rare presentations in the extremes of ages. The examination usually reveals a painless, round, well-circumscribed scrotal mass measuring up to 5 cm [3]. Adenomatoid tumors are most commonly located in the male genital tract, in the lower pole of the epididymis (tail), followed by the tunica albuginea and spermatic cord [5]. On ultrasound (US), these tumors typically appear as a solid, well-circumscribed mass.

In this case report, we discuss a possible case of adenomatoid tumor of the epididymis in an elderly man presenting with epididymo-orchitis and asymptomatic TRCC.

Case presentation

A 67-year-old Lebanese male patient presented to the emergency department at the American University of Beirut Medical Center, Lebanon, with a 10-day history of dysuria, urinary frequency, and urgency. The patient's medical history was notable for hypertension, benign prostate hypertrophy, hyperuricemia, and dyslipidemia. The patient was not on immunosuppressive treatment and did not have a history suggestive of immunologic problem. He was on amlodipine, valsartan, and bisoprolol for hypertension, as well as a statin for hyperlipidemia, and allopurinol. The patient did not have a significant surgical history. He does not smoke or consume alcohol. His father had coronary heart disease, and his mother had Parkinson disease. Personal and family histories were otherwise unremarkable, without testicular torsion or trauma. He has an administrative job and is married with children (Table 1).

On physical examination, the right scrotum was found to be three times larger than the left one, and the scrotal sac was erythematous. The exam also revealed a firm, non-tender intrascrotal mass, distinct from the testis. The mass looked to be arising directly from the right epididymis. The exam was otherwise normal.

Given his dysuria and other urinary symptoms, a urine culture was obtained. The culture revealed extended-spectrum beta-lactamase *Escherichia coli*. Ultrasonography showed a slightly disturbed echotexture of the right testicle with increased blood flow, and an associated septate reactive hydrocele with inflammatory changes. The US also revealed a 1.6 cm lesion within the right epididymal tail with some calcifications; this was reported as a possible adenomatoid tumor by the radiologist and the urology team (Figs. 1 and 2). Since adenomatoid tumor is

Table 1 Timeline showing pertinent procedures and findings

July 03, 2012: US of the kidneys and pelvis revealed left kidney heterogeneous hyperechoic lesion. Further evaluation (18 July 2012) by triphasic CT showed a Bosniak type 2 renal cyst.
10 May 2014: Admitted for urinary symptoms, enlarged scrotal sac.
12 May 2014: US of the testicles revealed right epididymo-orchitis with reactive inflammatory hydrocele. Lesion containing calcifications within the tail of the right epididymis possibly representing an adenomatoid tumor.
14 May 2014: US of the abdomen and pelvis revealed hyperechoic lesion within the superior aspect of the left kidney slightly increased in size. Further evaluation via a dedicated MRI examination recommended.
17 May 2014: MRI of the abdomen showed multiloculated cystic mass within the midpole of the left kidney. Cystic renal cell carcinoma was the most likely diagnosis.
6 June 2014: PET scan showed a well-defined hypodense lesion involving the anterior aspect of the left kidney measuring 4.3 × 3.3 cm and showing diffusely low activity (SUV_{max} 2.2) with a focus of increased activity along its superior anterior aspect with an SUV_{max} of 2.9 on the early phase and an SUV_{max} of 3.2 on the delayed phase.
19 November 2014: MRI the abdomen with gadolinium showed an interval increase in the size of the left kidney lesion measuring 4.7 × 5.2 cm (previously 3.9 × 4.7 cm). The mass again showed extension to the renal hilum.
21 November 2014: US of the testicles showed a decrease in the lesion in the right epididymal tail, showing some calcifications. This lesion can represent an adenomatoid tumor
Reactive right hydrocele and right small spermatocele or epididymal head cyst were noted.
2 April 2015: Partial left nephrectomy.
16 April 2015: Immunohistochemistry and microscopic findings revealed tubulo-cystic renal carcinoma with free surgical margins.
15 October 2024: Patient functional, assuming an advisorship position in a university.

PET, positron emission tomography; SUV_{max}, maximum standardized uptake value

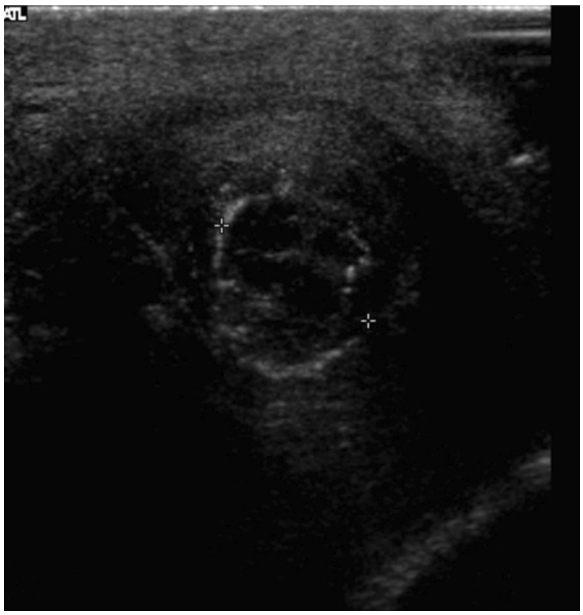


Fig. 1 Ultrasound showing a hypoechoic round paratesticular mass (two crosses indicate the margins)

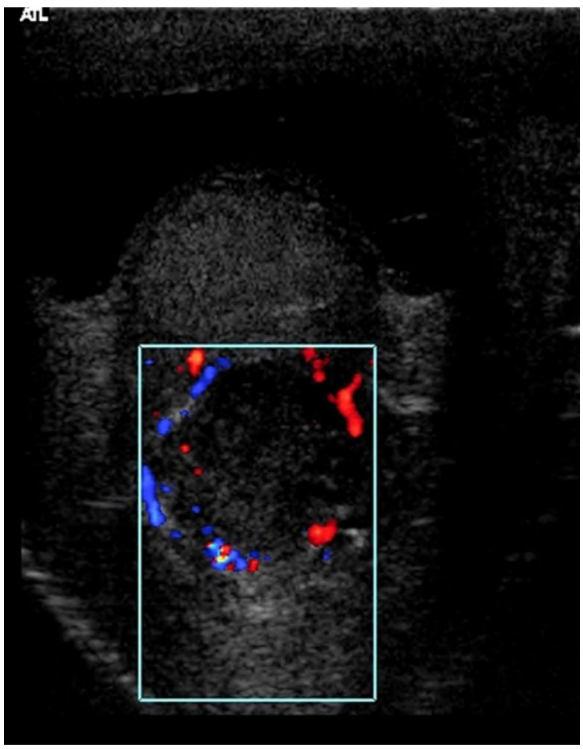


Fig. 2 Color Doppler ultrasound of the testicles shows the hypovascular nature of the lesion (lesion inside the rectangle)

a benign neoplasm with no malignant potential, no surgical intervention was taken.

Ultrasound of the abdomen revealed a hyperechoic mass within the superior aspect of the left kidney. This mass was seen on a CT 2 years prior to presentation and was reported by the radiologist to be consistent with a Bosniak type 2 cyst. MRI of the abdomen in May 2014 (Fig. 3) showed a multiloculated cystic mass within the midpole of the left kidney that may represent RCC.

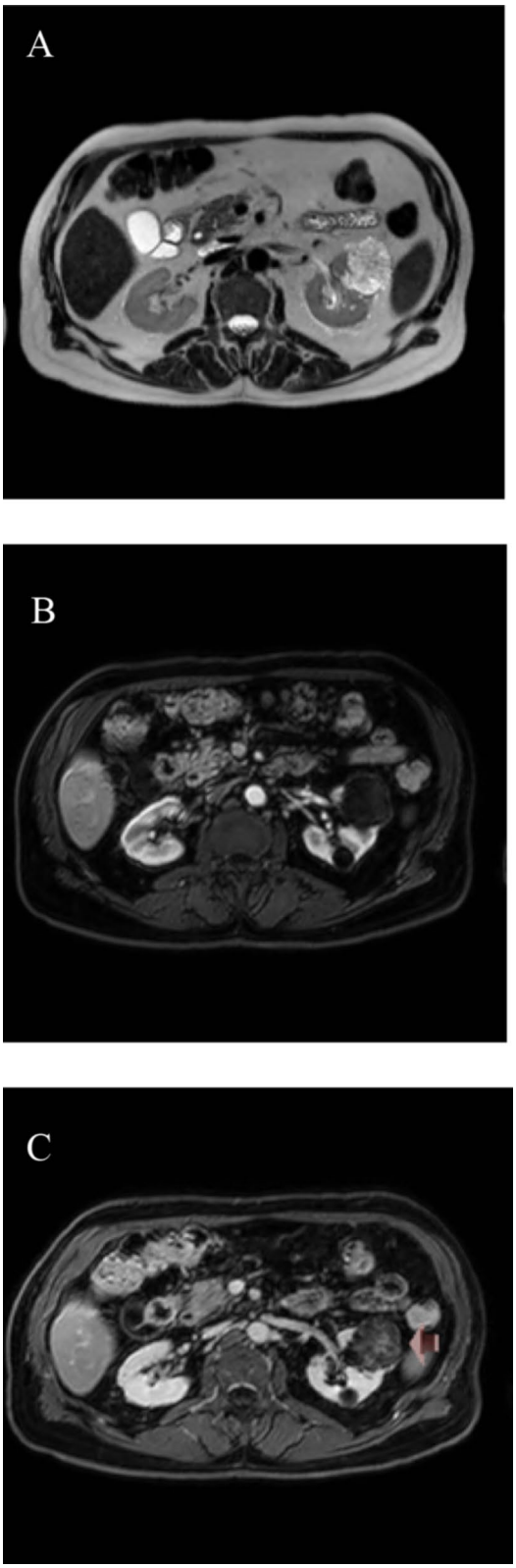
The patient was diagnosed with prostatitis and epididymo-orchitis. He was treated with piperacillin/tazobactam 4.5 g three times daily for 5 days and was discharged on trimethoprim/sulfamethoxazole double strength two tablets twice daily for 3 weeks.

Positron emission tomography (PET) scan (Fig. 4), done 3 weeks after the MRI on an outside basis did not report an epididymal lesion; however, a hypodense solid left kidney mass with a worrisome focal area of increased activity was noted.

MRI was repeated 6 months after discharge (November 2014) revealing an interval increase in the size of the left kidney lesion from 3.9×4.7 cm to 4.7×5.2 cm (Fig. 5). A decision for surgery was made, and the patient underwent left partial nephrectomy in April 2015. Histology confirmed the diagnosis of TRCC (Fig. 6). Laboratory studies including a complete blood count (CBC) and prostate-specific antigen (PSA) were repeated and were normal. Repeat testicular US (November 2014) showed complete regression of the right orchitis with partial regression of the epididymitis and a decrease in the right epididymal tail lesion with some calcifications. A total of 9 years after the surgery (October 2024), the patient is functional, with good health. The timeline for the major events and findings is shown in Table 1.

Discussions and conclusions

Ultrasound is the initial imaging modality for evaluating scrotal and testicular lesions. Adenomatoid tumors are solid and hypoechoic on ultrasound [3, 4]. Ultrasonography also allows a real-time maneuver where the testicle can be pushed by the sonographer, resulting in displacement of the testicle but not the lesion; this reveals the extratesticular location of the tumor [6]. Unlike usual benign lesions, these tumors may present with infiltration of adjacent tissues because they are not encapsulated [6]. In this case, it would be difficult to distinguish it from a malignant intratesticular tumor, and intraoperative histological analysis may become necessary to avoid



◀ **Fig. 3** Magnetic resonance imaging of left kidney lesion showing (**a**) heterogeneous predominantly high T2 signal; (**b**) no significant enhancement on the arterial phase, and (**c**) heterogeneous linear enhancement on the delayed images

unnecessary orchiectomy [7]. MRI is reserved for problem-solving; it usually demonstrates low T2-weighted signal intensity relative to testicular parenchyma and is better able to determine its exact paratesticular origin. Typically, they enhance slower and less than normal testicular parenchyma after gadolinium administration; however, this is variable [8].

Paratesticular tumors are rare, comprising less than 5% of all intrascrotal tumors. In 1945, Golden and Ash indicated for the first time that epididymal adenomatoid tumors are a common paratesticular neoplasm [9]. In 1976, Beccia *et al.* studied 314 epididymal tumors, of which 75% were benign. Of those, 73% were diagnosed as adenomatoid, followed by leiomyomas (11%), and papillary cystoadenomas of the epididymis (9%). Angiomas, lipomas, and hamartomas constituted the remaining 7% [10]. In our case, the diagnosis of adenomatoid tumor was a radiological one and was accepted by the urology team. Thus, the diagnosis is not definite.

Adenomatoid tumors arise more in the lower pole than the upper pole of the epididymis, by a ratio of 4:1. These tumors are usually unilateral and more common on the left side. Such tumors can also occur in the testicular tunica, testis, and spermatic cord and are rarely extratesticular. A total of 15–20% of cases are found to be associated with hydrocele [11]. These tumors are rare, slow-growing neoplasms, typically asymptomatic, with only 30% presenting with pain. In fact, around 5% of patients present with acute onset of inflammation and pain that suggest epididymitis [11]. Data are still contradictory regarding the histopathology of adenomatoid tumors, mostly favoring a mesothelial origin over a reaction to injury or inflammation [11]. There is an association between adenomatoid tumors and immunosuppression; this was not the case in our patient [12]. In our case, the presentation was unique, as the patient is an elderly man with a challenging diagnosis: from an acute prostatitis with urinary tract infection to an epididymo-orchitis with possible adenomatoid tumor associated with TRCC.

Regarding the TRCC, only 10% of the patients present with the classic triad of hematuria, flank pain, and abdominal mass [2]. Additionally, the growth in size of the cystic mass hides a malignant potential. In general,

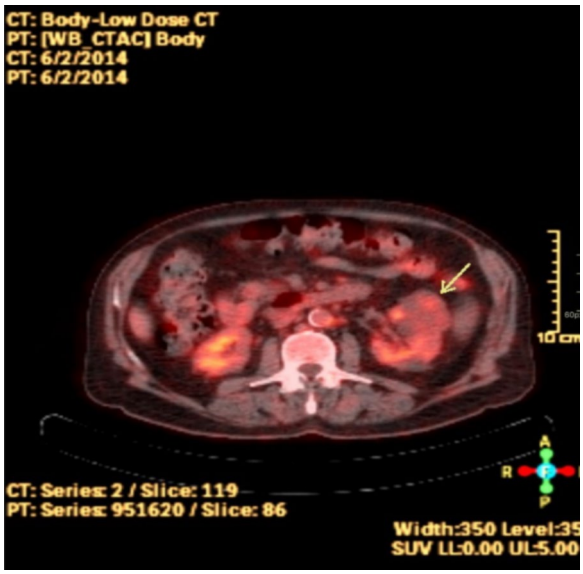
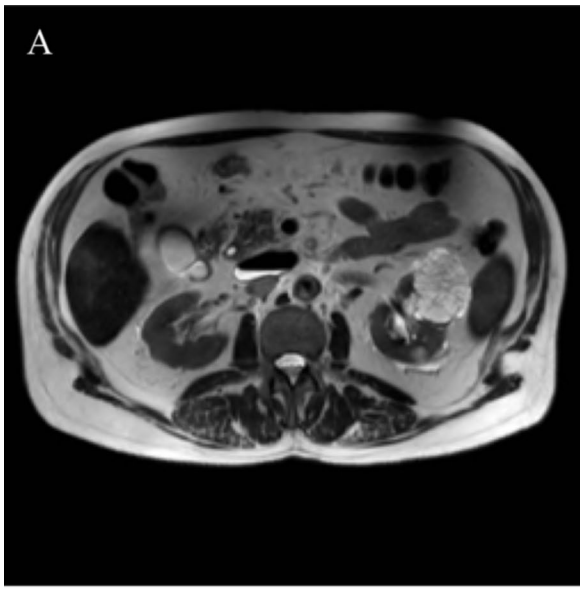


Fig. 4 Positron emission tomography (June 2014), arrow showing left renal mass



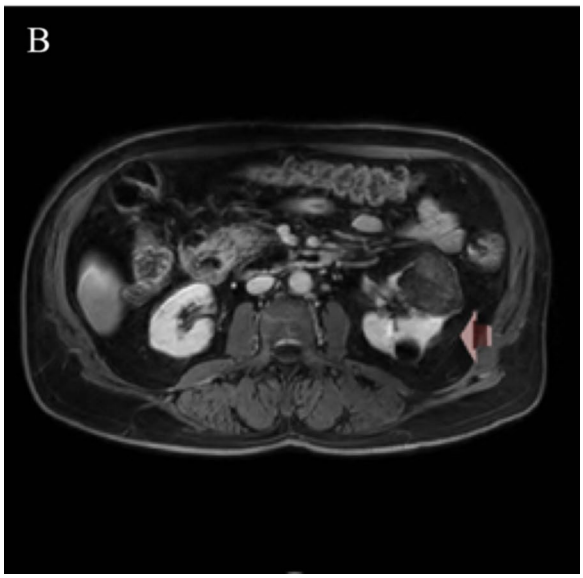


Fig. 5 Repeat magnetic resonance imaging after 6 months shows an increase in the size of the left kidney lesion on (**a**) a T2-weighted image and (**b**) delayed postcontrast gadolinium-enhanced images

such a tumor has a good prognosis and low rate of recurrence. This tumor may grow from Bosniak type II–IV cysts, as is the case of our patient [13]. Cystic RCC constitutes 10% of the presentations of renal cell carcinoma [14]. These may manifest as unilocular or multilocular. Definitive differentiation between cystic RCC and complex cysts is on pathologic examination [15]. Calcification, high attenuation (>20 HU) at unenhanced CT, signal intensity not typical of water on MRI, septations, loculations, enhancement, wall thickening, or nodularity are all criteria for classification as a complicated cyst/cystic renal mass [16].

In conclusion, TRCC, a rare type of RCC, might be the result of progression from Bosniak type II cysts. TRCC are usually treated by a nephron-sparing surgical approach. Adenomatoid tumors of the epididymis are rare tumors that may present at an older age. Physical examination and testicular US are important tools in the diagnosis. Benignity of adenomatoid tumor of the epididymis is reassuring and is a reason why it may usually be underreported.

Take-home messages

- Adenomatoid tumor of the epididymis is a rare asymptomatic benign tumor with no malignant potential that may occur in elderly man. Benignity of adenomatoid tumor of epididymis is reassuring and is a reason why it may usually be underreported.
- Ultrasound findings and follow-up can be enough for the presumptive diagnosis of adenomatoid tumor.
- Follow up with MRI to confirm possible US findings of RCC.

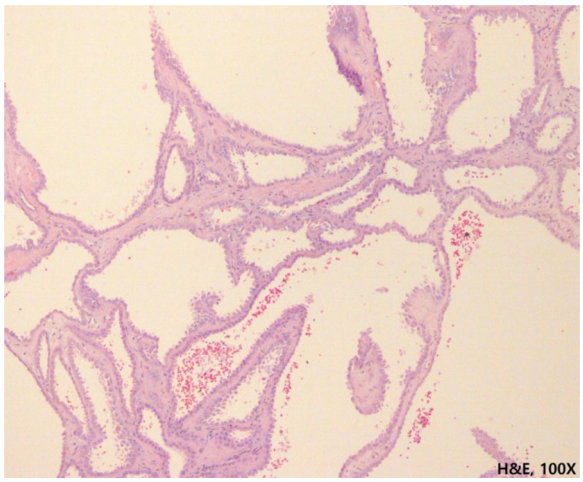


Fig. 6 Histology of the kidney tumor showing tubulocystic renal cell carcinoma

- Nephron-sparing surgery is optimal for TRCC considering the typically favorable histopathology.

Abbreviations

TRCC	Tubulocystic renal cell carcinoma
RCC	Renal cell carcinoma
US	Ultrasound
CT	Computed tomography
MRI	Magnetic resonance imaging
PET	Positron emission tomography

Supplementary Information

The online version contains supplementary material available at https://doi.org/10.1186/s13256-025-05165-7.

Supplementary Material 1.

Acknowledgements
We would like to acknowledge Dr. Mukbil Hourani for providing Fig. 4.

Author contributions
Literature review—Alexandra Ghadieh and Khaled El Khatib. Manuscript writing—Alexandra Ghadieh, and Bassem Saab. Clinical diagnosis and work up—Bassem Saab and Ghassan Hamadeh.

Funding
None.

Availability of data and material
Data were taken from patient's electronic medical chart after obtaining his consent.

Declarations

Ethics approval and consent to participate
According to the Institutional Research Board (IRB) policy at the American University of Beirut Medical Center, there is no need for IRB approval in case reports involving 1–3 subjects.

Consent for publication
Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing interests
No competing interests.

Received: 3 June 2024 Accepted: 11 February 2025
Published online: 20 March 2025

References

1. Chen S, Zhao J, Xin J, Yang X. Multimodal imaging findings of tubulocystic renal cell carcinoma: a case report. J Clin Imag Sci. 2024;14:37.
2. Zhang J, Liu B, Song N, Hua L, Wang Z, Gu M, Yin C. Diagnosis and treatment of cystic renal cell carcinoma. World J Surg Oncol. 2013;11:1–5.
3. Philips S, Nagar A, Dighe M, Vikram R, Sunnapwar A, Prasad S. Benign noncystic scrotal tumors and pseudotumors. Acta Radiol. 2012;53(1):102–11.
4. Benson CB, Doubilet PM, Richie JP. Sonography of the male genital tract. Am J Roentgenol. 1989;153(4):705–13.
5. Kim TJ, Kim SH, Sim JS, Seong CK, Lee DK. Ultrasonographic findings of an intratesticular adenomatoid tumor. J Ultrasound Med. 2000;19(3):227–9.
6. Amin W, Parwani AV. Adenomatoid tumor of testis. Clin Med Pathol. 2009. https://doi.org/10.4137/CPath.S3091.
7. Garriga V, Serrano A, Marin A, Medrano S, Roson N, Pruna X. US of the tunica vaginalis testis: anatomic relationships and pathologic conditions. Radiographics. 2009;29(7):2017–32.
8. Kim W, Rosen MA, Langer JE, Banner MP, Siegelman ES, Ramchandani P. US–MR imaging correlation in pathologic conditions of the scrotum. Radiographics. 2007;27(5):1239–53.
9. Golden A, Ash JE. Adenomatoid tumors of the genital tract. Am J Pathol. 1945;21(1):63.
10. Beccia DJ, Krane RJ, Olsson CA. Clinical management of non-testicular intrascrotal tumors. J Urol. 1976;116(4):476–9.
11. Akbar SA, Sayyed TA, Jafri SZ, Hasteh F, Neill JS. Multimodality imaging of paratesticular neoplasms and their rare mimics. Radiographics. 2003;23(6):1461–76.
12. Karpathiou G, Hiroshima K, Peoc'h M. Adenomatoid tumor: a review of pathology with focus on unusual presentations and sites, histogenesis, differential diagnosis, and molecular and clinical aspects with a historic overview of its description. Adv Anat Pathol. 2020;27(6):394–407.
13. Al-Jubouri AM, Khalil IA, Alhyari A, Alkabbani M, Badawi A, Goyal R, Al JK. Tubulocystic renal cell carcinoma: a case report of rare tumor. Radiol Case Rep. 2024;19(11):4836–40.
14. Hartman DS, Davis CJ Jr, Johns T, Goldman SM. Cystic renal cell carcinoma. Urology. 1986;28(2):145–53.
15. Murad T, Komaiko W, Oyasu R, Bauer K. Multilocular cystic renal cell carcinoma. Am J Clin Pathol. 1991;95(5):633–7.
16. Agnello F, Albano D, Micci G, Di Buono G, Agrusa A, Salvaggio G, Pardo S, Sparacia G, Bartolotta TV, Midiri M, Lagalla R. CT and MR imaging of cystic renal lesions. Insights Imaging. 2020;11:1–1.

Publisher's Note