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Case Report

Segmental testicular infarct with an associated testicular artery aneurysm: Case report of a rare clinical entity *

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ABSTRACT

Segmental testicular infarct is a rare clinical entity and can be a diagnostic challenge. Although cases are often idiopathic, underlying etiologies can include testicular torsion, epididymo-orchitis, trauma, vasculitis, and hypercoagulable states. Once suspected, an underlying testicular neoplasm should be excluded. We present a case of a 43-year-old male who developed acute onset left sided scrotal pain. A diagnostic scrotal ultrasound showed a focal, heterogeneous region in left testicle with absent focal Doppler signal, concerning for a segmental testicular infarction. There was no history of trauma, urinary symptoms, sexually transmitted diseases, or constitutional symptoms. Work up for associated underlying etiologies was negative. A computed tomography angiogram scan of the abdomen and pelvis revealed an incidental left testicular artery aneurysm. The patient's consulting multidisciplinary care teams included urology and vascular surgery. Urology deemed surgical intervention inappropriate for the segmental testicular infarct, and vascular surgery elected not to intervene on the testicular artery aneurysm due to risk of completing testicular infarct and damaging blood supply to the testis. The patient was discharged after achieving adequate pain control, and completion of inpatient work up. No underlying malignancy was diagnosed on follow up, and pain symptoms resolved. To the authors' knowledge, no literature exists describing the concurrent incidence of a segmental testicular infarct and an ipsilateral testicular artery aneurysm. In this report, we aim to further describe both diagnoses, and explore the association between the 2 entities.

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Introduction

Global testicular infarction, often secondary to testicular torsion, is a well-known clinical entity. On the contrary, segmental testicular infarction (STI), a rarity, presents a diagnostic challenge as it can mimic testicular torsion, tumor, or an intratesticular abscess. Literature on segmental testicular infarction is sparse [1–5]. Underlying etiologies of STI include partial torsion, acute epidiymo-orchitis, scrotal trauma, vasculitis, sickle cell disease, and hypercoagulable states [6,7]. Cases are, however, often idiopathic [8]. Diagnosis is established by scrotal ultrasound (US) and Doppler, and may be aided by Magnetic Resonance Imaging (MRI) [3,9].

Aneurysm of the testicular artery is exceedingly rare, with only a handful of cases described in literature [10–12]. These can be spontaneous, or secondary to trauma, infection, or inflammation, and can be a cause of acute scrotal pain [11,12].

The authors present a case of acute scrotal pain attributed to STI diagnosed on US, with a left testicular artery aneurysm (TAA) identified on further work up via computed tomography (CT) angiogram. To the authors' knowledge, no literature exists describing this concurrent phenomenon. The aim of this report is to describe the diagnoses of both the entities and explore the association between the TAA and the STI.

Case report

A 43-year-old male with no significant past medical history presented to the urgent care with acute onset of left scrotal pain. There was no history of trauma, urinary symptoms, sexually transmitted diseases, unintentional weight loss, fevers, chills, or night sweats. Physical exam demonstrated swollen left testicle with tenderness to palpation and manipulation. No obvious masses were noted on palpation. A diagnostic scrotal US performed showed a geographic, heterogeneous region in the superior pole of the left testicle with absent focal Doppler signal, concerning for a STI (Fig. 1). Patient was instructed for conservative management including nonsteroidal anti-inflammatory medications, supportive underwear, and icepack with referral to urology for follow up.

He presented to the emergency room (ER) two days later with worsening, severe left testicular pain. Work up in the ER showed normal electrolytes, complete blood count, coagulation profile, urinalysis, and microscopic urine studies. Repeat scrotal US demonstrated an increased area of STI (Fig. 2). The patient was admitted for pain control and further diagnostic studies to evaluate for underlying embolic event, vasculitis, and coagulopathy. Laboratory studies demonstrated mildly elevated ESR (erythrocyte sedimentation rate, 21 mm/Hr, normal 0-15 mm/h) and C-reactive protein (5.7 mg/dL, normal <0.5 mg/dL). Polymerase chain reaction testing for sexually transmitted disease including chlamydia and gonorrhoeae returned negative. Urine culture showed no microbial growth. Human immunodeficiency virus testing, and Syphilis screening were negative. Antiphospholipid antibody panel and immunologic testing including double-stranded DNA, antinuclear antibody screen, anti-neutrophil cytoplasmic antibody,



Fig. 1 – Grayscale and doppler ultrasound of bilateral testes. Focal, segmental, area of heterogenous reflectivity in the superior pole of the left testicle (white arrow) with absent focal doppler signal. Findings concerning for segmental testicular infarction



Fig. 2 – (A) grayscale and doppler ultrasound of bilateral testes, and (B) left testis superior pole ultrasound with doppler. Patient presented 48 hours later with worsening scrotal pain. Both images demonstrate increased size of left superior pole segmental testicular infarct (white arrows) compared to Fig. 1

C3 and C4 complements, myeloperoxidase antibody, (proteinase 3) antibody, scleroderma (Scl-70) antibody returned negative. Tumor marker alpha-fetoprotein was normal. Testing for Factor V Leiden mutation was negative.

A CT angiogram of the abdomen and pelvis was performed to evaluate for thrombosis and any vascular abnormality. This



Fig. 3 – (A) Coronal reconstructed CT maximum intensity projection (MIP) image show a left testicular artery aneurysm (white arrow); Three-dimensional segmented vascular model as seen in virtual reality (B) and (C) further illustrate the left testicular artery aneurysm (curved and straight white arrows, respectively)



Fig. 4 – Grayscale and doppler ultrasound of bilateral testes. Exam performed at 6 week follow up. Regressing size of segmental testicular infarct in the left superior/mid pole (white arrow). Patient's scrotal pain was resolved

revealed a 7 mm aneurysm of the left testicular artery (Fig. 3). Coronal maximum intensity projection (MIP) scrollable CT images demonstrating the left testicular artery aneurysm and its connection to the aorta are available in MP4 digital multimedia format file as online supplementary material. No additional vascular abnormality was identified. Given the risk of possible total infarct of the left testicle with embolization of the testicular artery, vascular surgery recommended active surveillance with repeat CT Angiogram. Surgical intervention was deemed inappropriate by Urology. Inpatient adequate pain control was achieved with a combination regimen of gabapentin, intravenous Toradol, acetaminophen, and hydromorphone. The patient was subsequently discharged with instructions for follow up with Urology and Vascular Surgery.

Sonographic follow up after 6 weeks showed a smaller area of left superior pole testicular infarct (Fig. 4). The patient was clinically asymptomatic; there was no scrotal pain, urinary symptoms, or hematuria.

Discussion

STI is a rare clinical entity. In a series by Bilagi et al, out of 8091 patients undergoing testicular US over a period of 6 years, 24 (0.3%) met criteria for STI [5]. In a smaller series of 342 patients with scrotal diseases queried over a period of 6 years, Fernandez-Perez found an incidence of 3.5% [3].

Patients with STI present with acute scrotal pain. The age distribution is later than the classic testicular infarction/testicular torsion and is seen in the 2nd-fourth decades of life. Most cases are idiopathic, although associations with epididymo-orchitis, intermittent testicular torsion, vasculitis, sickle cell disease, hypercoagulable states, trauma, polyarteritis nodosa, diabetic microangiopathy, embolic disease, and previous surgery (hernia repair, varicocelectomy, vasectomy, cysto-prostatectomy) have been documented [5–7,13–18]. Interestingly, an extensive work up of our patient was negative for these associated conditions.

The primary diagnostic tool for evaluation is US. Grayscale findings demonstrate a round, elongated, or wedge-shaped peripheral region of mixed reflectivity, which evolves into a hypoechoic lesion with an echogenic rim [3–5]. Color Doppler shows absent or markedly decreased flow in the focal area. Previous case series have described a predilection for the upper pole [3–5,13], as was seen in our case.

Magnetic resonance imaging (MRI) is a useful adjunct tool in evaluating segmental testicular infarction. Lesions are focal geographic, or wedge shaped and are isointense on T1 weighted imaging (WI) and hypointense on T2WI. There may be rim-enhancement on post-contrast T1WI [3,5].

Differential diagnoses include testicular neoplasm and abscess. Gradual regression on follow-up US may help exclude a neoplasm. Further, testicular neoplasms usually show normal or elevated color Doppler signals. Recognition of US findings, with possible aid of MRI, clinical history, and tumor markers, can prevent an unnecessary orchiectomy. Focal orchitis will demonstrate increased Doppler signal. Clinical history and signs of infection can help distinguish an abscess.

The testicular arteries branch off the abdominal aorta, pass through the deep inguinal ring into the spermatic cord, and enter the scrotum via the inguinal canal. Each testicular artery subdivides into anterior/superior and posterior/inferior divisions, penetrates the tunica albuginea, travels through the tunica vasculosa, and then further subdivides into centripetal branches and intertubular arterioles supplying the lobules containing the seminiferous tubules [19]. The posterior/inferior divisions share anastomoses with the deferential and cremasteric arteries. This anatomical configuration may explain the preferential distribution of SIT in the upper pole due to relative lack of collateral blood supply.

Aneurysm of the testicular artery is exceedingly rare. An extensive search of literature revealed less than a handful of such cases [10-12]. Interestingly, an association between TAA and STI has not been described. Previously documented cases of TAA have suggested a spontaneous etiology [10-12]. In our patient, etiology is likely spontaneous versus congenital, given the absence of trauma and infection. Diagnosis can be made with US and Doppler examination, CT angiogram, or MRI angiogram. Management is usually conservative. Surgical excision or endovascular treatment can be performed for symptomatic or enlarging aneurysms [12]. In our patient, the vascular surgery team elected conservative management of the TAA. Previous case reports have described a rare risk of total testicular infarct following endovascular intervention for abdominal aortic and aorto-iliac aneurysms [20-23]. Although no such literature exists describing risk of total testicular infarct following endovascular intervention in the testicular artery, the vascular surgery team at our institution elected conservative, nonoperative management to mitigate the risk of total testicular infarction with endovascular intervention.

To the authors' knowledge, no case has been described of a STI and a concurrent TAA. These two findings may be coincidental or associated. The authors hypothesize that turbulent flow dynamics from the TAA may have led to STI via micro-embolic phenomenon. Hemodynamics of small vessel aneurysms has been extensively studied. Numerous studies have investigated the association between the contributing factors as turbulent flow, irregular flow patterns, wall sheer stress, and endothelial dysfunction and the formation of an aneurysm and its rupture [24,25]. Parameters of hemodynamic stress including temporal flow patterns, inflow and mean velocities, vortex formation, wall sheer stress, among others, have also been related to endothelial homeostasis and development of atherosclerotic lesions and intra-luminal thrombi [25-29]. Further, the thrombi associated with a peripheral arterial aneurysm have been shown to predominantly complicate with distal microemboli [30]. These studies lend credibility to associating the hemodynamics of the left TAA with micro-emboli that may have contributed to STI in our patient.

Limitations of our case report include a lack of pathologic diagnosis of testicular infarct (due to ethical reasons). Further, hemodynamic studies of aneurysms have largely been studied in the abdominal aorta, intracranial aneurysms, and to a lesser degree in peripheral arterial aneurysms as referenced above. Due to the rarity of TAA, there are not dedicated hemodynamic studies of aneurysms at this location. Interestingly, studies for intracranial aneurysm hemodynamics share a similar diameter range (5–10 mm) to the TAA noted in this case (7 mm), perhaps allowing for some relative comparison of hemodynamics when accounting for size, even though the microenvironments are different.

Conclusion

In conclusion, both STI and TAA are rare diagnoses. The radiologist plays a pivotal role in diagnosing STI, which is identified on ultrasound as round or wedge-shaped lesion with focal decreased blood flow. MRI may aid in diagnosis. Management is conservative and follow up US should be performed in 6–12 weeks to rule out underlying malignancy. TAA can be idiopathic/spontaneous, congenital, or post-traumatic. The authors hypothesize that micro-emboli from local turbulent flow dynamics from the ipsilateral TAA may have contributed to the STI. TAA are likely to be found incidentally, and diagnosis can be established by CT Angiogram. No standard of treatment exists due to the rarity of the condition. In this case, vascular surgery elected nonoperative management.

Supplementary multimedia: coronal reconstructed maximum intensity projection images (mp4 digitial multimedia video) show origin of the left testicular artery from the aorta, and the left testicular artery aneurysm.

Patient consent

Informed consent was obtained from the patient for publication of the case report and for all imaging studies. The consent was uploaded to our institution's electronic medical record.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.radcr.2022.02.068.

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