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LETTER TO THE EDITOR

Testicular angioleiomyoma presenting with haematospermia

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Dear editors,

We report an exceedingly rare case of primary testicular angioleiomyoma to increase awareness of this rare entity among urologists and pathologists. To our knowledge, only one case of this rare testicular tumour has been reported in the literature to date, and this is the first report of its presentation with haematospermia.[1]

A 65-year-old patient was admitted to our clinic for the evaluation of painless haematospermia. The patient's past medical history was unremarkable. At presentation and during the scrotal and testis examination, there was no palpable mass. The patient had no voiding symptoms, and there were no signs of infection. The patient also denied experiencing any recent trauma. A digital rectal examination of the prostate was normal. The ultrasonographic findings disclosed a well-circumscribed ovoid hypoechoic nodule within the right testis, near its upper pole, measuring approximately 0.5 cm, and moderate hydrocele (**Figure 1a**). The values of serum prostate-specific antigen, alpha-fetoprotein, beta-human chorionic gonadotropin and lactate dehydrogenase were normal.

The patient underwent an elective exploration of the right testis through an inguinal incision. An intratesticular tumour was excised and sent for pathological examination. A frozen section analysis was performed, and a diagnosis of a benign leiomyomatous tumour was established. The tumour was completely excised, and the testis was preserved. A definitive pathological diagnosis was made on paraffin-embedded sections of formalin-fixed tissues. Microscopically, the tumour was composed of a well-circumscribed proliferation of smooth muscle bundles that surrounded thick-walled blood vessels with partially patent lumina (**Figure 1b**). The smooth muscle cells showed no atypia or mitoses. Immunohistochemically, the perivascular proliferating smooth muscle cells and the muscular wall of the blood vessels showed immunoreactivity for smooth muscle antigen and desmin. It was determined that this was a case of classic angioleiomyoma by observation of microscopic appearances and immunohistochemical analysis. The patient experienced an uneventful recovery and has had no signs of haematospermia to date.

Benign smooth muscle tumours are classified histologically as leiomyomas and angioleiomyomas.[2] Angioleiomyoma is a benign tumour arising from the vascular smooth muscle, and they typically present between the third and fifth decades of life.[3] These tumours commonly occur in the subcutis of the extremities and, to a lesser extent, of the head and trunk.[2] Angioleiomyoma presents as a painful mass in approximately 60% of cases.[3]

Like other smooth muscle tumours, angioleiomyoma can theoretically arise anywhere in the body from tissues containing smooth muscle. Within the scrotum, structures such as the epididymis, spermatic cord, tunica albuginea, tunica vaginalis testis and blood vessels could be sites of origin of these benign tumours.[1] The origin of intratesticular angioleiomyoma is still controversial. Possible sites of origin include the smooth muscle cells of the vascular tree and contractile cells in the tunica propria of the seminiferous tubules.[4,5]

Benign smooth muscle tumours are extremely rare in the testes. Intratesticular leiomyomas have been reported only a few times in the English literature.[4,5] To our knowledge, intratesticular angioleiomyoma has been reported only once, by Lavis *et al.*[1] The mode of presentation is typically a testicular mass with or without pain, and most cases are treated with radical orchiectomy. The small tumour in our case is unique in that it manifested with haematospermia, and no palpable mass or pain was present.

Haematospermia is often considered to be a symptom of little significance, and its aetiology is poorly understood. In most cases, it is caused by inflammation of the prostate and seminal vesicles, but in a small percentage of men, it may be a manifestation of genitourinary malignancy, particularly prostate cancer or testicular cancer.[6,7] The current recommendations state that an initial investigation should include a focused history, clinical examination, digital rectal examination, serum prostate-specific antigen, urine cultures and screening for sexually transmitted diseases but not scrotal ultrasonography.[6] Recently, an algorithm for haematospermia put forth by the American Academy of Family Physicians recommended scrotal examination and ultrasonography to exclude infection and testicular cancer.[8] Our patient had no history of prostate disease, infections or systemic diseases that could explain the haematospermia, making the angioleiomyoma the most likely cause of the bleeding. The discovery of a benign tumour in this case may only be incidental, but scrotal ultrasonography is easy and noninvasive and should be performed in all patients presenting with haematospermia.

Any solid mass within the scrotum is considered malignant until proven otherwise. The differential diagnosis includes inflammation (i.e., orchitis), abscess and neoplasm. Other intratesticular neoplasms that have a similar appearance to angioleiomyoma include malignant

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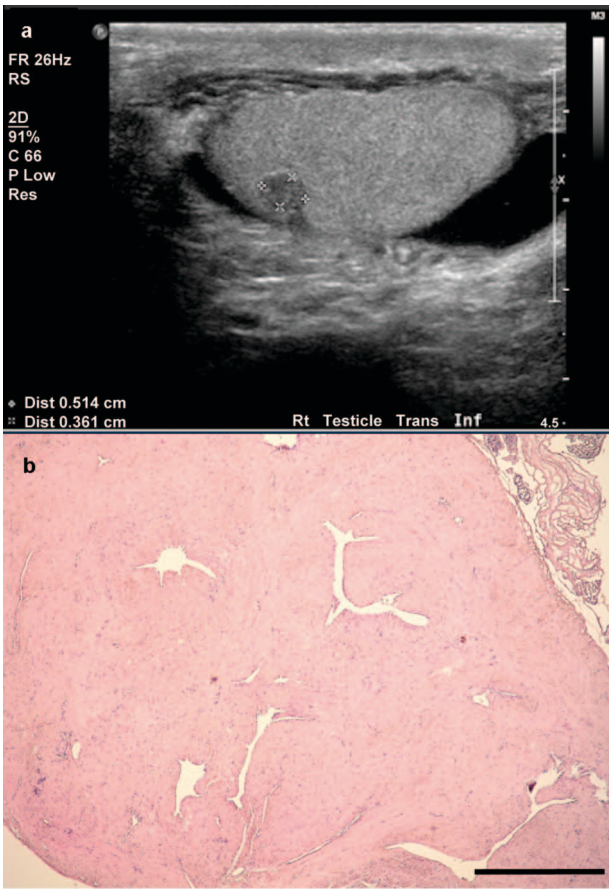

Figure 1 (**a**) Ultrasonographic image of the right testis showing a small hypoechoic inhomogeneous lesion measuring 0.5 cm and moderate hydrocele. (**b**) The tumour was composed of a well-circumscribed proliferation of smooth muscle bundles that surrounded thick-walled blood vessels with partially patent lumina. Scale bar = 700 μm.

tumours (e.g., lymphoma and germ cell tumours), tumours with malignant potential (e.g., Leydig cell tumours and Sertoli cell tumours) and benign tumours. Benign testicular tumours are rare, and the most common are fibroma, haemangioma, lipoma, angiomatoid tumours and epidermoid and dermoid cysts.[4] Because there is no reliable imaging technique to clinically distinguish benign lesions from the more common malignant tumours, we chose to perform testis preservation surgery with a frozen section analysis. Two studies have evaluated the accuracy of frozen section examination in testicular tumours.[8,9] Both studies demonstrated that frozen section examinations could accurately differentiate benign from malignant testicular tumours. Based on these data, a frozen section examination should be included in the management of testicular masses of unknown origin.

We believe that it is important to be aware that benign intratesticular lesions do exist and that the threshold for surgical excision should be low. Accurate assessment of an undetermined testicular tumour should facilitate organ preservation if the benign nature of the mass is detected preoperatively or during surgery. This case also underscores the importance of scrotal examination and ultrasonography in patients presenting with haematospermia.

AUTHOR CONTRIBUTIONS

GS performed the clinical diagnostics and surgery in the patient. GS and AD retrieved the clinical data and images. DT performed the frozen section analysis. DT and AD established the pathological diagnosis. GS and AD wrote the draft of the manuscript, which was revised by DT and BK. DT and BK took part in the discussion of the case. All authors have read and approved the final manuscript.

COMPETING FINANCIAL INTERESTS

The authors declare no competing financial interest.

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