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ORIGINAL ARTICLE

Multiparametric sonographic imaging of a capillary hemangioma of the testis: appearances on gray-scale, color Doppler, contrast-enhanced ultrasound and strain elastography

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Abstract We report a case of a lobular capillary hemangioma in a 66-year-old man, who presented with left testicular pain, with an asymptomatic incidental right testicular lesion found on ultrasonography. The sonographic examination demonstrated a heterogeneous mainly isoechoic intratesticular lesion with marked vascularity on the color Doppler examination. Further evaluation with contrast-enhanced ultrasound and strain elastography was performed; the multiparametric imaging suggested a benign tumor. The multidisciplinary team decision with patient consent was to perform a radical orchiectomy with subsequent histopathology confirming a benign lobular capillary hemangioma.

Keywords Testis · Ultrasonography · Contrast-enhanced ultrasound (CEUS) · Strain elastography (SE) · Capillary haemangioma

Riassunto Riportiamo un caso di angioma lobulare capillare in un uomo di 66 anni, presentatosi con dolore testicolare sinistro e con all'ecografia una lesione incidentale al testicolo di destra. L'esame ecografico ha rilevato una lesione eterogenea, principalmente isoecogena, con florida vascolarizzazione al color Doppler. Abbiamo proceduto ad ulteriore valutazione con ecografia con mezzo di contrasto ed elastografia, e l'imaging multiparametrico ha suggerito un tumore benigno. In accordo con il paziente, il team multidisciplinare ha deciso di procedere ad una orchidectomia radicale e l'esame istopatologico ha confermato un angioma lobulare capillare benigno.

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Introduction

Testicular neoplasms that are derived from connective tissue, blood vessels and musculature are uncommon and intra-testicular tumors of vascular origin are extremely rare; both are benign in nature [1–7]. Testicular hemangioma typically occurs in patients younger than 20 years, the age in which a primary germ cell tumor of the testis may present, necessitating a radical approach to management with orchidectomy [8]. The differentiation between a benign and malignant lesion of the testis is crucial, but not always possible on imaging techniques currently deployed. Although malignant lesions of the testis are overwhelmingly more common, prevention of an orchidectomy in the presence of benign disease is desirable [9]. We present a case of an intratesticular capillary-type hemangioma in an adult that mimicked a primary testicular malignancy on baseline and color Doppler ultrasonography, but where the addition of contrast-enhanced ultrasound (CEUS) and strain elastography (SE) imaging (multiparametric ultrasound imaging) identified the lesion as benign, allowing for potential conservative focal partial surgical excision.

Case report

A 66-year-old man, who presented with one month history of left-sided mild scrotal pain, was referred for a sonographic examination of the scrotum by the examining Urologist. The patient was well with no relevant medical or surgical history, except for a smoking history of 35 years and medically treated atrial fibrillation. Ultrasonography, using a Acuson Siemens 3000 (Mountain View, CA, USA) and a 9L4 multi-frequency linear transducer, demonstrated an incidental, asymptomatic, right-sided well-circumscribed testicular mass measuring approximately 22 × 16 mm in the central aspect of the right testis (Fig. 1a). This was of slightly increased echogenicity (relative to normal testis) with lobulated margins, otherwise homogeneous echo structure, and evidence of hypervascularity on color Doppler examination (Fig. 1b), features which initially suggested the presence of a neoplastic lesion, possibly a primary germ cell tumor or lymphoma. To further characterize the lesion, a CEUS examination was performed (CPS™, Siemens, CA), administrating 4.8 mL of ultrasound contrast (SonoVue™, Bracco, Milan, Italy) via a peripheral vein, followed by 10 mL of 0.9 % saline injection, following the established protocol for a CEUS of the testis [10, 11]. After injection of contrast, the lesion demonstrated an early and avid arterial peripheral nodular enhancement, with central contrast filling in the venous/parenchymal phase, with evidence of wash out at 90 s following contrast administration (Fig. 2a–d), but with persistent remaining hyper enhancement compared to the background testis parenchyma. Strain elastography (SE) was also performed on Hitachi HV900 (Hitachi Medical Corporation, Tokyo, Japan) using a 14–6 MHz linear transducer and Hitachi real-time tissue elastography (H-RTE™). A visual score of 3 was estimated, based on a scoring system previously reported [12, 13] and a mean strain ratio of 2.3 calculated, suggestive of a soft lesion (Fig. 3). Multiparametric imaging findings combining the B-Mode, color Doppler, CEUS and SE examinations were suggestive of a benign tumor. Laboratory tests, including serum levels of relevant tumor markers, particularly beta-human chorionic gonadotrophin (beta-HCG = 2U/L; normal, <5 U/L), alpha-fetoprotein (AFP = 2U/L; normal <7 U/L), LDH (LDH = 222 U/L, normal <240 U/L) were within normal limits; a normal white blood cell count was recorded (WBC = 7.11/mmc, normal 4.000–10.000/mmc). The case was discussed in the Urology multidisciplinary meeting and, even though the sonographic characteristics were suggestive of a benign mass, the surgeons suggested a radical right inguinal orchiectomy as the size of the lesion made partial orchidectomy impractical. The patient accepted this course of management and underwent a right orchidectomy 10 days following the sonographic examination. Histological examination demonstrated a well-circumscribed hemorrhagic tumor within the center of the testicular parenchyma. The lesion had a pushing border, implying a lesion that is expansile, i.e., has the potential to grow, but it was not infiltrative into the adjacent normal tissue like a malignant process, and showed no involvement of the rete testis or epididymis. The lesion had a lobular architecture composed of multiple capillaries, features of a lobular capillary hemangioma (Fig. 4). No cause was identified for the left testicular pain, a common complaint, which was managed conservatively.

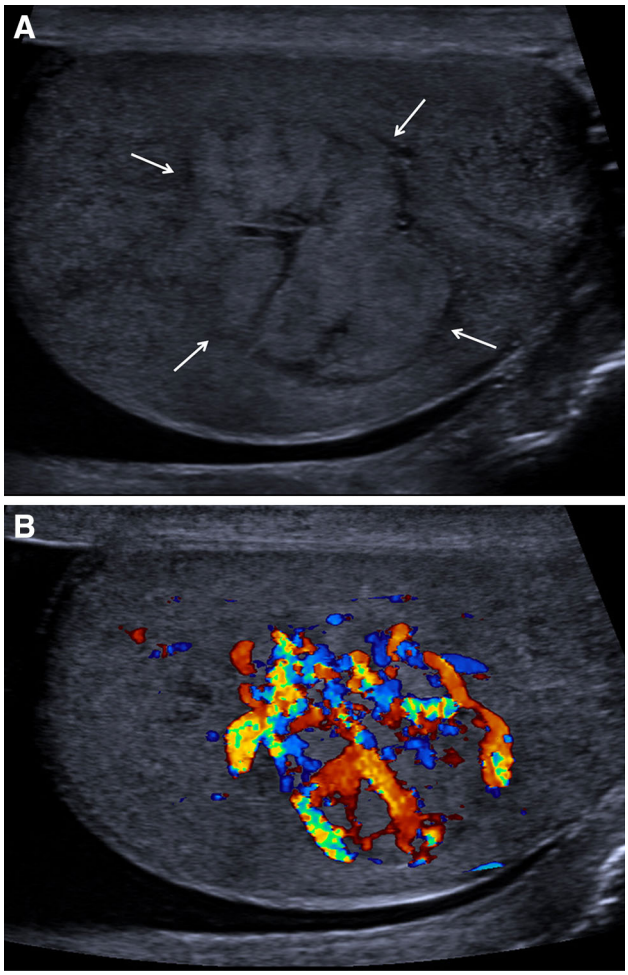

Fig. 1 **a** A lobulated iso-echoic lesion (*arrows*) within the central aspect of the right testis, well demarcated from the testicular parenchyma. **b** There is a marked increase in color Doppler flow to the lesion in comparison to the surrounding normal testicular parenchyma

Discussion

A hemangioma is the most common soft tissue tumor; testicular occurrence is extremely rare with most testicular hemangiomas present in infancy, childhood, and young adults [4, 7]. A total of 43 cases of testicular hemangiomata

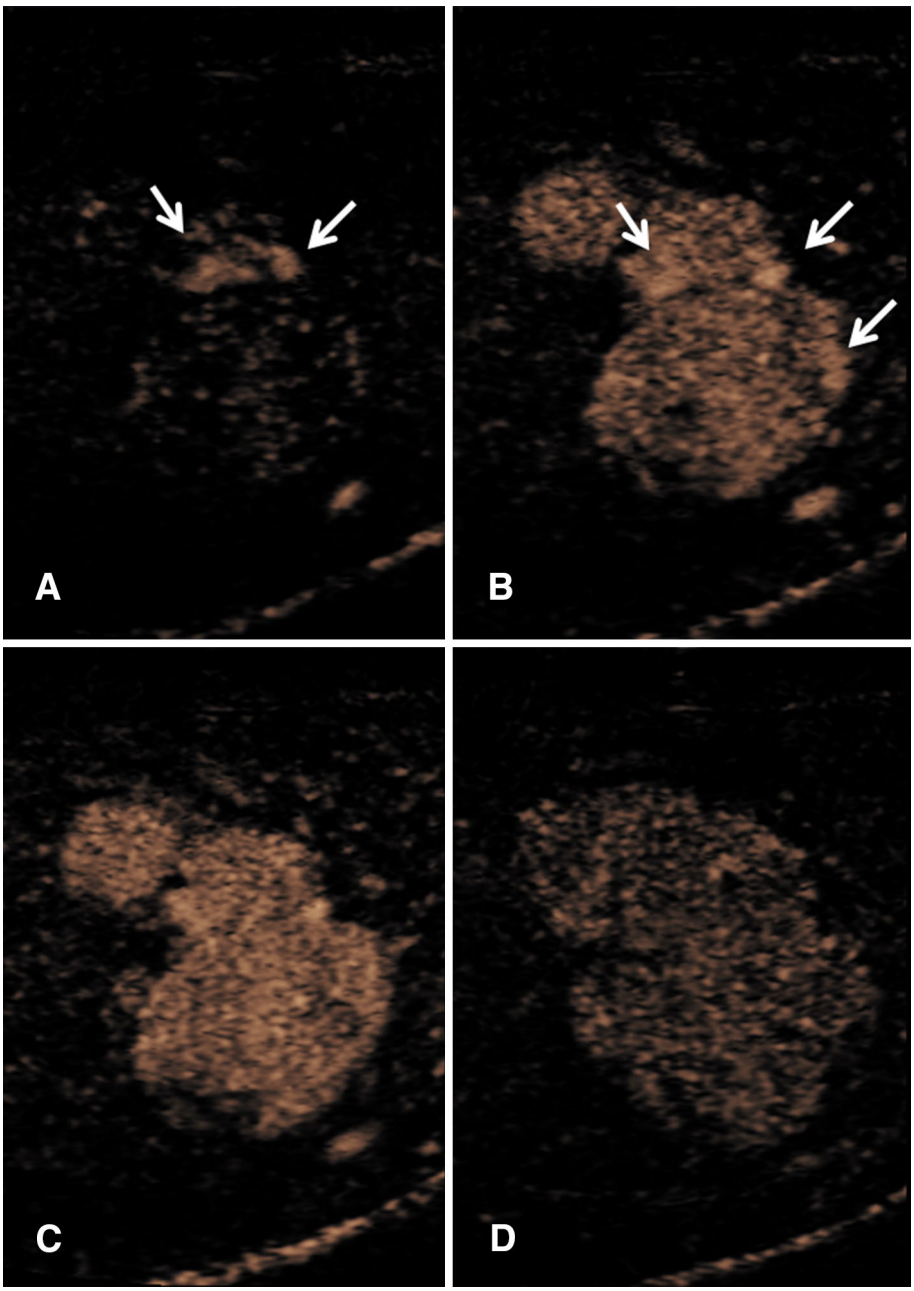

Fig. 2 **a** Following the administration of SonoVue™, as contrast, at 28 s following administration, there is "globular" enhancement (*arrows*) over the upper aspect of the lesion. **b** At 35 s, the lesion is enhancing much more avidly than that of the surrounding testicular parenchyma, with the peripheral "globular" enhancement (*arrows*) still evident. **c** At 40 s, the enhancement remains much more prominent in the lesion but the peripheral "globular" enhancement is less obvious. **d** At 60 s enhancement of the lesion has diminished but is still more prominent than the surrounding testicular enhancement

have been documented, comprising three types (cavernous, capillary, and epithelioid), without any previous reports of the use of multi-parametric ultrasound imaging, particularly using the newer techniques of CEUS or SE examination [1–7, 14]. Several previous reports have described the gray-scale and color Doppler appearances of capillary hemangiomas of the testis [4–7, 14]. The predominant appearance on gray-scale imaging is of a hypo-echoic area, with increased color Doppler flow, and invariably this is a capillary hemangioma on histology [4]. When a more complex or heterogeneous appearance is documented, a cavernous hemangioma is reported [4].

Ultrasonography is established as the first-line imaging method for testicular symptoms, and adequately identifies abnormalities to allow for appropriate clinical management. The finding of a focal intra-testicular lesion nearly always implies the presence of a malignant lesion, and orchidectomy is the standard surgical management. With the advent of more sophisticated sonographic techniques, the ability to ascertain further information with regard to the vascularity and the texture of the tumor has progressed with a number of groups claiming more detailed interpretation of the findings using CEUS and SE [9, 10, 12, 15, 16]. The prospect of testicular sparing surgery in the pre-

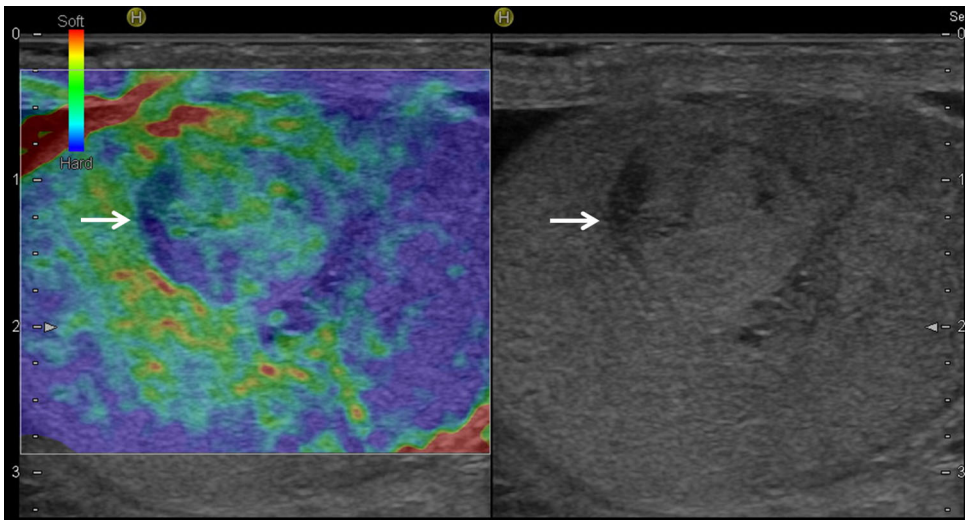

Fig. 3 The paired strain elastography images demonstrate the "soft" nature of the lesion (*arrow*) with a predominant *green color*, but with demarcation from the surrounding testicular parenchyma

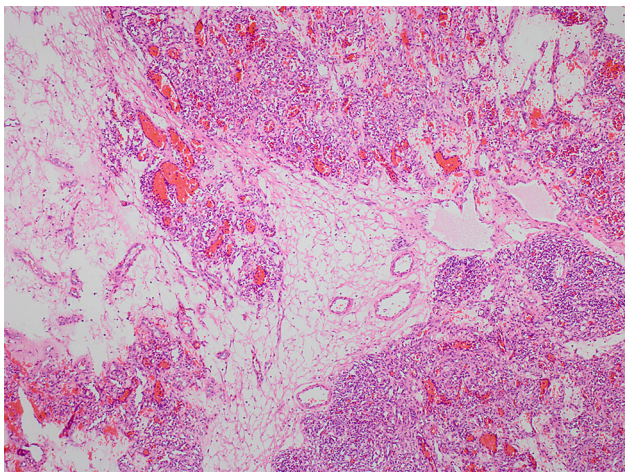

Fig. 4 H+E section, low power view (×10 magnification). A rich capillary network arranged in variably sized lobules with an intervening edematous septal stroma

operative knowledge of a benign intra-testicular lesion is desirable [17]. CEUS enables a substantial improvement in the accuracy of sonography in the differential diagnosis of small testicular nodules [10], and in the present case CEUS demonstrates a unique pattern of contrast enhancement (peripheral nodular pattern) that, in retrospect following histology, mimics the CEUS appearances of hemangioma in the liver and spleen [11, 18]. Elastography, an ultrasound measure of the stiffness of tissue, could potentially identify the "hard" lesion as more likely malignant and the "soft" lesion benign [15], although there are exceptions to this with the benign epidermoid lesion characteristically "hard" on SE [10, 12]. In the present case, SE identified the lesion as soft, potentially benign, and allowing the observers to suggest a benign abnormality on the presentation of sonographic examination. Importantly, these sonographic techniques are much less expensive and more convenient than magnetic resonance (MR) imaging, which is often the "fallback" imaging technique with uncertain grey scale and color Doppler sonographic findings [19].

Previous reports of the sonographic appearances of a hemangioma of the testis are sparse, and there are no previous reports with the additional use of CEUS and SE appearances. Previously imaged lesions, predominantly <10 mm in size, have demonstrated hypoechogenicity, and increased vascularity on color Doppler examination [5–7, 14]. Larger lesions have been described as heterogeneous [4]. The lesion reported here was larger (>22 mm) and iso-echoic to the testis with increased vascularity on color Doppler examination. Hemangioma elsewhere assumes an echogenic appearance, and in the testis this may be size dependent with the smaller lesion being echo-poor, and the large lesions more echogenic.

These findings are relevant for the individualized treatment of patients [20]. The addition of CEUS and SE has added to the armamentarium to increase operator confidence; patients with testicular lesions may be monitored with close follow-up and not be submitted to operation immediately. Awareness on the part of the operator is essential for the correct interpretation and diagnosis. In dealing with a testicular mass in a patient with no serum elevation of beta-human chorionic gonadotropin and alpha-fetoprotein, and an ultrasound study showing a vascularized mass, this neoplasm, although rare, should be considered in the differential diagnosis.

Compliance with ethical standards

Conflict of interest (Silvia Bernardo MD, Eleni Konstantatou MD, Dean Y. Huang MRCP FRCR, Annamaria Deganello MD, Marianna Philippidou BSc MBBS FRCPath, Christian Brown BSc MD FRCS (Urol), Maria E. Sellars MBBS FRCR, Paul S. Sidhu MRCP FRCR) declare that they have no conflict of interest.

Ethical standard All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2000 (5).

Informed consent All patients provided written informed consent to enrollment in the study and to the inclusion in this article of information that could potentially lead to their identification.

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