Case Report

Multiparametric Ultrasound (mpUS) of a Rare Testicular Capillary Hemangioma

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Capillary hemangioma is a rare entity among testicular tumors. We demonstrate the case of an 18-year-old patient with palpatoric and sonographic conspicuous left testicle and negative serum tumor markers (α-fetoprotein, β-human chorionic gonadotropin, and lactate dehydrogenase). Ultrasound (US) imaging represented an isoechogenic lesion with high vascularization in both power Doppler and microflow imaging with central feeding artery. Both strain elastography and shear wave elastography demonstrated a stiff lesion compared to surrounding testicular tissue. While contrast-enhanced ultrasound (CEUS) clearly depicted high vascular load, time intensity curve (TIC) analysis was able to show shorter median transit time, higher peak enhancement, and higher wash-in area under the curve compared to regular testicular tissue. Histopathological examination revealed a lobular constructed and rich vascularized proliferation without cellular atypia and feeder vessels with positive reaction to CD34, CD31, CD99, and Vimentin. Proliferative activity was quantified to 3–5% by Ki-67 index. Two days after surgery, the patient could leave the hospital in subjective wellbeing. While histology remains the gold standard to make a precise diagnosis of capillary hemangiomas due to small case numbers and variety of this benign tumor, the combination of multiparametric US and clinical information may be a promising future tool in preoperative assessment.

1. Introduction

Capillary hemangioma is a rare entity among testicular tumors. Nevertheless, this benign vascular tumor is an important differential diagnosis since the most common malignant neoplasm in young men is testicular carcinoma, with increasing incidence [1]. Testicular capillary hemangioma seems to appear especially in young men as well, whereby this assumption should be considered with caution because of the small number of reported cases [2]. In addition to capillary form of testicular hemangioma, there are cavernous and epithelioid variants described as well. Multiparametric ultrasound (mpUS) means the combination of the established methods B-mode US, color-coded duplex sonography (CCDS), contrast-enhanced US (CEUS), and elastography [3]. Elastography measures the stiffness of tissue and can be distinguished in strain (longitudinal pressure generated by the examiner’s compression of tissue beneath the probe) and shear wave elastography (SWE; impulse of the probe generating a transversal wave which velocity relates to local stiffness). CEUS uses the dynamic real-time character to depict vascularization and perfusion of a certain tissue or lesion in comparison to the surrounding tissue by evaluating wash-in and wash-out of a contrast agent. Overall, a variety of different sonographic parameters (multiparametric) can give in synopsis the diagnosis and may decrease operator dependency by providing quantitative results [3].

2. Case Description

We demonstrate the case of an 18-year-old patient who was admitted to our interdisciplinary US centre by an
external urologist with palpatoric and sonographic conspicuous left testicle. Clinical examination revealed a caudal induration of the left testicle, which was nevertheless indolent. Epididymis and contralateral testicular parenchyma were inconspicuous. Serum tumor markers were negative: α-fetoprotein (α-AFP, 1.1 ng/ml), β-human chorionic gonadotropin (β-HCG, <0.1 U/l), and lactate dehydrogenase (LDH, 172 U/l).

B-mode ultrasound of the left testicle showed an isoechogenic, round-shaped lesion in the central left testicle up to 11 mm diameter with a hypoechogenic peripheral rim (Figure 1(a)). Power Doppler imaging (PDI) and microflow imaging (e.g., SMI (superb microvascular imaging)) depicted hypervascularization that clarified the finding of a vascular tumor (Figures 1(b) and 1(c)). While PDI only demonstrated strong vascularization, SMI was able to show bigger feeder vessels in the central part of the lesion with high density of smaller central vessels around (Figure 1(c)). Strain elastography demonstrated a harder lesion compared to the surrounding testicular tissue, while the lesion had a higher stiffness (up to 6.5 m/s) in 2D SWE compared to regular testicular tissue (0.5 m/s) (Figure 2). Furthermore, stiffness of the lesion was less homogenous than the regular surrounding tissue.

CEUS examination was performed and interpreted by a single high-experienced radiologist with more than fifteen years’ experience in CEUS (EFSUMB level 3) using a high-end ultrasound system (Apio i500, Canon, Otawara, Japan) with a linear broadband transducer (i14L5; Canon, Otawara, Japan). B-mode US was optimized using spatial compounding, frequency-based compounding, ApliPure™ level 5, differential Tissue Harmonic Imaging (dTHI)®, and Precision Imaging© with level 4 Speckle Reduction (SR). CEUS was performed at 9 MHz, 10 fps configured with a very low MI (0.07) to avoid early microbubble destruction. A bolus of 2.4 ml of ultrasound contrast agent (SonoVue®, Bracco Imaging, Milan, Italy) was injected up to three times.
The hypervascularized lesion showed early and strong contrast enhancement compared to the surrounding tissue. Arrival time imaging depicted a shorter arrival time feeder vessel with short central filling of the lesion due to high vessel density (Figure 3). In comparison to the surrounding tissue, time intensity curve (TIC, Table 1) measurement showed a clearly higher peak intensity (45.6 vs. 3.4), shorter mean transit time (7.0 vs. 10.1 sec), and higher wash-in area (121.3 vs. 9.1), while time to peak showed no difference (4.4 vs. 4.2 sec) (Figure 4).

The patient received enucleation and intraoperative incision by highly experienced surgeons. Histopathological examination revealed a lobular constructed and rich vascularized proliferation without cellular atypia and feeder vessels. Immunohistology revealed positive reaction to CD34, CD31, CD99, and Vimentin. Moreover, there was a significant capillary growth form in the tumor’s periphery between

<table>
<thead>
<tr>
<th>TIC parameter</th>
<th>Hemangioma</th>
<th>Testicular tissue</th>
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<tbody>
<tr>
<td>PI (1.0E-5 AU)</td>
<td>45.6</td>
<td>3.4</td>
</tr>
<tr>
<td>TTP (s)</td>
<td>4.2</td>
<td>4.4</td>
</tr>
<tr>
<td>MTT (s)</td>
<td>7.0</td>
<td>10.1</td>
</tr>
<tr>
<td>Slope (1.0E-5 AU/s)</td>
<td>13.0</td>
<td>1.0</td>
</tr>
<tr>
<td>Area (1.0E-5 AU/s)</td>
<td>2007.9</td>
<td>130.0</td>
</tr>
<tr>
<td>AreaWI (1.0E-5 AU·s)</td>
<td>121.3</td>
<td>9.7</td>
</tr>
<tr>
<td>AreaWO (1.0E-5 AU·s)</td>
<td>1886.6</td>
<td>120.3</td>
</tr>
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</table>

ROI within the capillary hemangioma demonstrated higher PI, shorter MTT, higher slope, and higher wash-in/wash-out rates compared to regular testicular tissue. Abbreviations: ROI: region of interest; PI: peak intensity; TTP: time to peak; MTT: mean transit time; AreaWI: area wash-in; AreaWO: area wash-out.
preexisting testicular tubules. The capillaries were continu-
ously endowed with pericytes and showed partial expression 
of WT-1. Proliferative activity was quantified to 3–5% by 
Ki-67 index. Two days after surgery, the patient could leave 
the hospital in subjective wellbeing.

3. Discussion

The high vascularization in our case could have been shown 
congruous with PDI, SMI, and CEUS. Since hyperenhancement 
of a testicular lesion had a positive predictive value of 97.4% for 
neoplasia [4], the sonographic findings in our case indicated 
potential malignancy. Besides, the sonographic findings with 
central feeding vessels could allow a differentiation to benign 
Leydig cell tumor, which is suggested by a short filling time or 
by a circumferential vessel with a rapid centripetal filling [5]. 
The wash-in of the hemangioma was higher and more rapid 
than the surrounding tissue, as demonstrated by a shorter time 
to peak and higher peak enhancement and wash-in rate. The 
homogenous enhancement could be thought to be due to a high 
blood flow velocity and an increased microvessel density, while 
tumors like seminomas or burn-out tumors often show areas of 
less enhancement due to necrosis [6]. Furthermore, malignant 
lesions were more often described to be hypoechogenic com-
pared to the surrounding tissue [7]. Contrary to our results, cap-
illary hemangioma has also been demonstrated as a 
hypoechogenic lesion with low signal in CCDS [8].

In general, CEUS can be a very helpful method to investi-
gate microvascularization of testicular lesions. CEUS may 
be valuable in the assessment of contrast enhancement in 
small intratesticular masses (<5 mm) where color-coded 
duplex sonography comes to its limits because small testicu-
lar tumors may appear avascular. Since it is possible to distin-
guish solid and cystic or burn-out tumors, previous studies 
found no key finding in enhancement pattern to differentiate 
between benign and malignant tumors especially in small 
lesion (<10 mm). Furthermore, clinical presentation may be 
similar between testicular hemangioma and malignant testicu-
lar lesions with slight testicular pressure leading to pain.

CEUS has advantages over contrast-enhanced magnetic 
resonance imaging (MRI) including unmatched temporal 
resolution due to continuous real-time imaging [9]. US 
remains the primary imaging method in unclear testicular 
pain or incidental finding of testicular lesion. Therefore, 
CEUS is a fast and easy tool to elucidate unclear B-mode 
findings. Moreover, it is a safe tool because CEUS examina-
tions do not use radiation and the contrast agent applied 
has no renal, thyroid, or cardiac toxicity.

While orchietomy is recommended as the method of 
choice, enucleation and intraoperative incision showed a 
benign stroma tumor in this case, so the rest of the organ 
could remain. Nevertheless, histology remains the gold stan-
dard to make a precise diagnosis of a testicular tumor. Due to 
small case numbers and variety of this benign tumor, the 
combination of contrast-enhanced ultrasound, elastography, 
and clinical information (tumor markers) may be a promis-
ing future tool for treatment planning to avoid unnecessary 
orchietomy especially in young patients.
Conflicts of Interest

None of the authors reports a relationship with industry and other relevant entities—financial or otherwise—that might pose a conflict of interest in connection with the submitted article.

References


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