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A great malignancy mimicker: Testicular epidermoid cysts with atypical sonographic and MRI appearance

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ABSTRACT

Testicular epidermoid cyst (TEC) is an uncommon benign testicular lesion that can be successfully cured via lesion enucleation and at the same time preserving patient's fertility. Doppler ultrasound (US), contrastenhanced MRI and tumor markers are helpful in the diagnosis of TEC if the lesion does not show typical characteristics such as onion skin and target appearance. Herein, we report a case of TEC without typical structural characteristics but heterogeneously mixed echogenic content in US examination, no internal vascularity in the color Doppler study and lack of contrast enhancement in MRI images. These additional findings are helpful for diagnosing TEC.

Introduction

Testicular epidermoid cysts (TEC) is a rare, benign, slow-growing tumor that can be cured by testis-sparing surgery.¹ However, TEC with atypical image appearance mimicking malignancy, leading to unnecessary radical orchiectomy, highlights the importance of identifying the unconventional characteristics of TEC in order to differentiate between remediable cysts and cancerous lesions. Herein, we report a patient with testicular tumor presenting atypical US and MRI findings, who received orchiectomy. However, the lesion was later proven to be TEC pathologically.

Case presentation

A 22-year-old male came to hospital due to a gradually enlarging right scrotal nodule for one year. Physical examination showed a 1.5 cm \times 1.5 cm soft, non-tender nodule in the right testis with no lymphade-nopathy over the groin area. Tumor marker levels, including hCG and

AFP, were within normal limits. US examination showed a well-defined 1.4 cm \times 1.6 cm hypoechogenic intra-testicular nodule in the right testis with an irregular margin, heterogeneously mixed echogenic content as well as hypoechogenic cystic components, and a few partial mural echogenic reflectors (Fig. 1A). In MRI examination, this lobulated nodule demonstrated an irregular border, homogeneously low-signal intensity on T1WI and heterogeneously iso- to low-signal intensity on fat-suppressed T2WI (Fig. 1B and C). In addition, soft-tissue enhancement was not found on contrast-enhanced T1WI (Fig. 1D). After preoperative assessment, right radical orchiectomy was performed under the request of the patient. The final histopathology revealed right TEC with stratified squamous epithelium lining, filling with keratin (Fig. 2A and B).

Discussion

TEC is a rare benign tumor, accounting for 1–2% of testicular masses. 1 They are most commonly seen in Caucasian men between the

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Fig. 1. US and MRI Images of TEC.

(A) Sagittal grayscale US images of the right testis shows heterogeneous hypoechoic with hyperechoic foci (arrow) and acoustic shadow (B) T2- weighted MRI shows an ill-defined heterogeneously iso- to low-signal intensity lesion (C) T1- weighted MRI shows low-signal intensity without clear margin (arrow) (D) Contrastenhanced T1WI shows a heterogeneously low-signal intensity lesion without soft tissue enhancement.



Fig. 2. Pathological Image of Atypical TEC.

(A) Gross photograph of sectioned specimen, demonstrating smooth and well-defined lesion filled with white-yellow paste-like material (B) Microscopic visualization (H&E stain, $40 \times$) revealed that the cyst was lined by flattened squamous epithelium (arrow) and filled with laminated keratin in the center (asterisk). Normal seminiferous tubules are present beside the cyst (arrowhead). (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

age of 20 and 40. However, the exact etiology and biological behavior of TEC was a matter of dispute since it was reported.² Pathological features of TEC include: (1) a cystic lesion in the parenchyma of the testis; (2) an outer fibrous capsule wall with calcified epithelial lining; (3) inner laminated rings with more compact keratin debris in the center of the cyst. These traits can be of a striking resemblance to malignant

neoplasms.

Clinically, US is used as the primary image tool to evaluate testicular masses. Typical sonographic features of TEC include mural echogenic reflector; alternating hyperechogenic and hypoechogenic rings, known as typical onion skin appearance; and a hypoechoic rim with a hyperechoic center, known as bull's eye or target appearance.² TECs on MRI

can also present target sign or bull's eye sign.¹ The low signal intensity rim is produced by a fibrous capsule and epithelial lining; the high signal intensity mid area is desquamated keratin debris with high water and lipid content; the low signal intensity center is attributable to the dense debris. Regardless of the anatomical imaging characteristics, TEC usually shows neither internal flow on Doppler sonography, nor contrast enhancement on MRI. Conversely, intra-testicular neoplasms always display early rapid contrast enhancement on MRI. Thus, the absence of vascularity and contrast enhancement can be a hint to differentiate TEC from testicular malignancy.

In our case, no target sign or onion skin, no internal vascularity and no contrast enhancement were demonstrated on Doppler sonography and MRI, respectively. US study revealed an irregular and partially indistinct border, heterogeneous and partially hypoechogenic content with some focal or linear hyperechogenicity at the peripheral area while irregular margin with heterogeneously low signal intensity content were observed on T2WI of MRI study. These atypical image appearances might be attributable to the uneven and asymmetrical accumulation of keratin debris and desquamated epithelium. The uneven distribution of keratin also blocked the formation of inner laminated rings content, resulting in the irregular border, lobulated content and the absence of onion skin appearance on US and MRI images.

Actually, malignant testicular tumors can be overlapping with atypical appearances of TEC, as shown in our case. This increases the difficulty of making correct diagnosis. On US, seminoma typically shows features of well-defined, homogenously hypoechogenic content, while non-seminomatous tumors appear as an ill-defined heterogeneous lesion with cystic components and calcifications.³

Apart from structural evaluation, tumor marker can be a valuable indicator of the testicular cancer. For non-seminomatous germ cell tumors, elevation of AFP and hCG are 52%–72% and 30%–63%,

respectively.⁴ As for TEC with typical or atypical imaging, tumor markers are always within normal limits.^{1,2}

Conclusion

If a testicular tumor with normal tumor markers presents a lobulated contour, heterogeneously mixed echogenic appearance in the US examination, no internal vascularity in the color Doppler study and lack of contrast enhancement in MRI images, TEC should be taken into the list of differential diagnosis.

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Declaration of competing interest

None.

References

- Cho JH, et al. Sonographic and MR Imaging findings of testicular epidermoid cysts. Am J Roentgenol. 2002;178(3):743–748.
- Manning MA, Woodward PJ. Testicular epidermoid cysts sonographic features with clinicopathologic correlation. J Ultrasound Med. 2010;29(5):831–837.
- Marth D, Scheidegger J, Studer UE. Ultrasonography of testicular tumors. Urol Int. 1990;45(4):237–240.
- 4. Dieckmann KP, et al. Serum tumour markers in testicular germ cell tumours: frequencies of elevated levels and extents of marker elevation are significantly associated with clinical parameters and with response to treatment. *BioMed Res Int.* 2019;2019:5030349.