

Concomitant bilateral testicular epidermoid cysts

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ABSTRACT

يعتبر الكيس الجلدي في الخصيتين ورماً حميداً نادراً ما يحدث، ويصيب 1-2% من جميع كتل الخصية الحميدة التي تم استئصالها. تم الإبلاغ عن حوالي 300 حالة حتى تاريخنا هذا. كما تم الإبلاغ في الغالب عن الإصابة في الجانب الواحد في الأدب الإنجليزي. ولكن، يعتبر الظهور في كلتا الجانبين نادراً جداً، هنالك ثلاثة تقارير سابقة تم الإبلاغ عنها للأكياس الجلدية في كلتا الخصيتين. حقيقة أنها بالكامل مسؤولة عن العلاج بواسطة الاستئصال الموضعي، من أجل إنقاذ المريض من استئصال الخصية. تعد معرفة سمات التصوير بالموجات فوق الصوتية الخصائية مهمة جداً لتجنب التدخل العلاجي غير الضروري. نقدم هنا حالة للكيس الجلدي في كلتا الخصيتين التي تسمح نتائج الفحص بالموجات فوق الصوتية (US) الخصائية باستئصال الجزء المصاب من الخصية بدلاً عن الاستئصال الكامل للخصية.

Epidermoid cyst of the testis is a rare benign germ cell tumor, comprising 1-2% of all resected benign testicular masses. Approximately 300 cases have been reported to date. Unilateral involvement has often been reported in the English literature. However, bilateral occurrence is very rare and to the best of our knowledge, there are only 3 previous reports of bilateral testicular epidermoid cysts. The fact that they are completely benign makes them amenable to treatment by local excision, thereby saving patient from orchidectomy. Recognition of their characteristic ultrasonographic features is very important to avoid unnecessary intervention. We present here, a case of bilateral epidermoid cyst in which characteristic ultrasound (US) findings allowed testis-sparing enucleation instead of radical orchidectomy.

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Epidermoid cysts (EC) though common benign epithelial tumors of brain and meninges, have also been reported at several other locations including the spleen, salivary gland, and pancreas, as well as in the testis. Testicular epidermoid cysts constitute 1% of all intratesticular tumors.^{1,2} The purpose of present case report is to highlight the characteristic sonographic findings of epidermoid cysts, which should alert the radiologist to the benign nature of the lesion, allowing the surgeon to plan testis sparing surgery rather than a radical orchidectomy.

Case Reports. A 19-year-old Bahraini male who had presented with bilateral testicular pain and discomfort following a recent mild trauma to the scrotum, was referred to us for ultrasound (US) examination of scrotum. Ultrasound examination using Advanced Technology Laboratories (HDI5000) 7-12MHz linear array transducer, revealed 1.5 x 2.25 cm well defined hypoechoic spherical lesion containing few echogenic foci and hyperechoic capsule in the central portion of right testicle (Figure 1) with absence of color flow on Doppler US in the lesion (Figure 2). Similarly, the left testicle showed approximately 1.95 x 1.89 cm size isoechoic avascular lesion surrounded by a thick rim of alternating hypo and isoechoic layers (Figures 3 & 4). The surrounding testicular parenchyma was normal. His past history revealed a similar complaint 3 years earlier for which he underwent US examination. A similar finding had been noted in the previous US and a diagnosis of hematoma was suggested at that time due to the history of trauma. On comparison with previous US, no interval change in size and the appearance of the bilateral testicular lesions was seen. His laboratory test results including serum β -HCG and α -fetoprotein were normal. Based on characteristic US findings and negative tumor marker results, a diagnosis of benign bilateral testicular mass consistent with epidermoid cyst was suggested. The physical examination revealed no evidence of lymphadenopathy or gynecomastia. He underwent operative frozen section biopsy followed by excision of the masses and sparing of the testicle. Pathological examination revealed cyst



Figure 1 - Grey scale image of right testicle showing a hypoechoic lesion with echogenic foci within it and hyperechoic capsule around.

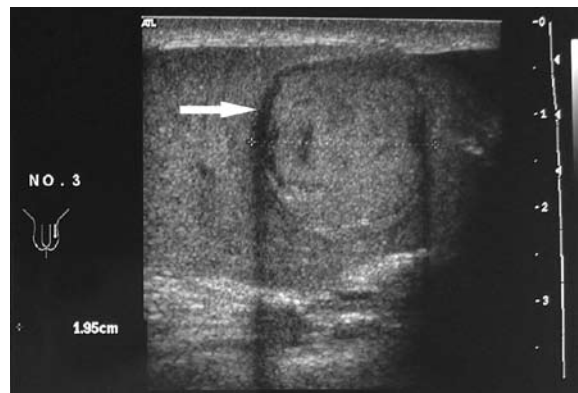


Figure 3 - Grey scale image of left testicle showing an isoechoic lesion with few small hypoechoic foci within and surrounded by an alternating hypo and isoechoic rim.



Figure 2 - Color Doppler image of right testicle showing the hypoechoic lesion to be avascular.

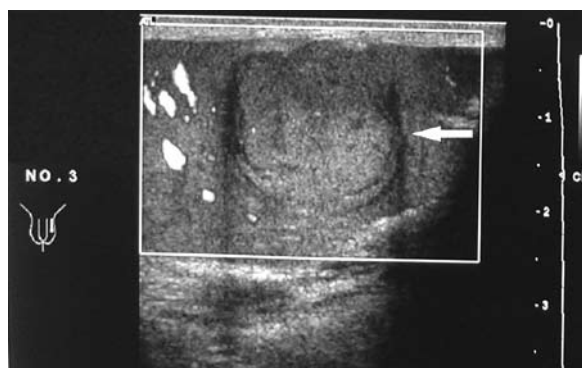


Figure 4 - Color Doppler image of left testicle showing lack of vascularity in the isoechoic lesion.

containing keratin and wall composed of fibrous tissue lined by squamous epithelium diagnostic of testicular epidermoid. His postoperative course was uneventful.

Discussion. Epidermoid cyst (also known as keratocyst) of the testis, first described in 1942 by Dockerty and Priestley,³ is a rare benign tumor, accounting for 1-2% of all testicular tumors.³⁻⁶ The reported cases have occurred in white and Asian persons. One series reports an incidence of 8% in a Chinese population.⁷ Clinical differentiation of epidermoid cyst from other testicular tumors is difficult. A typical presentation is a painless nodule incidentally discovered during self-examination or routine physical examination. Scrotal pain, scrotal enlargement, or vague discomfort are some other infrequently occurring clinical complaints. Presentation is usually in the second to fourth decades of life, however, any age group can be affected. The lesions usually measure 2-3 cm on average with a slight (56%) higher incidence to the right.⁴⁻⁸ Of approximately 300 reported cases in the literature,

only 3 had bilateral involvement.^{5,9,10} The histogenesis of epidermoid cyst is unknown. However, their origin from the monodermal development of teratoma has been suggested. Teratoma of the testis however, may have the elements of ectodermal, mesodermal, and endodermal layers.^{4,5} Price's¹ established widely accepted criteria of epidermoid cyst: (a) the lesion must be an intraparenchymal cyst; (b) the lesion must contain keratin or amorphous material; (c) the cyst wall should contain fibrous tissue with a complete or incomplete inner lining of squamous epithelium; (d) the cyst must contain no teratomatous components (for example, sebaceous gland, hair follicles) within the cyst wall or within the parenchyma of testis, and (e) no scar may be seen in the remaining testicular parenchyma.¹ The presence of teratomatous components and a parenchymal scar signify a burnt-out malignant germ cell tumor. Although, sonographic appearance of testicular epidermoid cysts may be variable, certain features have been identified, which suggest the histological diagnosis.⁶ They show a well-circumscribed

hypoechoic, avascular mass with a hyperechoic rim corresponding to dense fibrous capsule. Any calcification in the capsule exhibits a typical hyperechogenicity with acoustic shadowing. Sometimes concentric rings of alternating hypo and hyperechogenicity are seen within the cyst known as “onion-ring” appearance. This corresponds to the alternating layers of compacted keratin and loosely arranged desquamated squamous cells on the histological examination. An echogenic center may be seen within the hypoechoic lesion known as “target” or “bull’s” appearance, which corresponds to the histologic finding of innermost keratinized debris formed centrally.^{1,8,11} Recognition of these characteristic ultrasonographic features by radiologist is very important as they suggest the benign nature of the lesion.⁷ Our case showed a hypoechoic avascular mass with few echogenic foci with a hyperechoic rim around in the right testicle, while isoechoic avascular mass with alternating hypo and hyperechoic rim in left testicle. However, because of the considerable overlap in sonographic characteristics of epidermoid cyst and malignant masses, the role of sonography in exclusion of testicular malignancy is limited.⁸ Recently, MR imaging has been used to investigate testicular lesions. On MRI, peripheral low-signal-intensity zone is seen on both T1 and T2-weighted images due to the outer fibrous capsule, epithelial lining, and adjacent compact keratin. The center composed of dense debris and calcification, also produced low-signal, while high-signal intensity between 2 low-signal-intensity areas are due to desquamated cellular debris containing both water and lipid content correspond to the alternating hyperechogenicity or alternating echogenicity seen at US.¹¹

Management of testicular epidermoid cyst is a debatable issue. Inguinal orchiectomy is the standard

treatment for intratesticular masses suspected to be malignant neoplasms. However, based on characteristic imaging findings coupled with benign clinical picture, negative tumor markers, and careful frozen section examination, testicle-sparing surgery, or enucleation has been practiced more often recently.^{3,4,8}

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