

CASE REPORT

Bilateral Testicular Epidermoid Cysts

Norman Loberant, Shweta Bhatt¹, Edward Messing², Vikram S. Dogra¹

Department of Radiology, Western Galilee Hospital, Israel, ¹Departments of Radiology and ²Urology, University of Rochester, NY, USA

Address for correspondence:

Dr. Vikram S. Dogra,
601 Elmwood Avenue, University of
Rochester, NY - 14642, USA.

E-mail:

vikram_dogra@urmc.rochester.edu

Received : 13-09-2010

Accepted : 25-09-2010

Published : 01-01-2011

DOI : 10.4103/2156-7514.73502

ABSTRACT

Testicular epidermoid cysts are the most common benign tumors of the testes, but account for only 1–2% of all testicular tumors. In a young man presenting with a testicular mass, a high index of suspicion must be maintained for the malignant testicular germ cell tumor, which is 50-times more common than testicular epidermoid cyst. Bilateral testicular epidermoid cysts are a very rare condition, with only a few reports in the literature. It is extremely important in this condition to make a correct pre-operative diagnosis on imaging to enable a testis-sparing surgery.

Key words: Congenital, epidermoids, ultrasound, testes

INTRODUCTION

Testicular epidermoid cysts are the most common benign tumors of the testes, but account for only 1–2% of all testicular tumors. We present the case of a young man with bilateral testicular epidermoid cysts (TECs) with atypical features and review the imaging, management, and controversies concerning this entity.

CASE REPORT

A 17-year-old male presented to the Ultrasound Department for evaluation of a palpable mass in the left testis. An ultrasound examination demonstrated bilateral heterogeneous avascular testicular masses. The right testicular mass measured 1.5 cm × 1.3 cm × 1.2 cm and demonstrated a somewhat concentric, lamellated appearance with alternate hypoechoic and echogenic layers, known as the “onion skin appearance” [Figure 1]. The left testicular mass was multilobular and had a heterogeneous appearance. It measured 1.9 cm × 1.7 cm × 1.2 cm [Figure 2]. Tumor markers, including alpha-fetoprotein (AFP) and beta human chorionic gonadotropin (β-HCG) were within normal limits. In order to further determine the etiology of these lesions, the patient underwent an open biopsy of the left testicular

lesion. A frozen section of this lesion showed it to be an uncomplicated epidermoid cyst. The right testicular lesion was not biopsied or removed.

DISCUSSION

In a young man presenting with a testicular mass, one must maintain a high index of suspicion for malignant testicular germ cell tumor, which is 50-times more common than testicular epidermoid cyst, the most common benign testicular tumor.^[1,2] TEC accounts for only 1–2% of all testicular tumors and generally presents in the same manner (painless scrotal mass) and in the same age group (20–40 years) as malignant neoplasms. In three published studies, the average age of patients was between 22 and 24 years.^[2-4] TEC also presents in pediatric patients, comprising 3% of the testicular tumors in children.^[2]

The histogenesis of TEC is not entirely elucidated. One concept is that TEC represents a monodermal variety of teratoma, with the presence of ectoderm only; however, the consistent lack of testicular intraepithelial neoplasia (carcinoma *in situ*) in the adjacent parenchyma has cast doubts on this theory.^[1] Pathologic examination in TEC usually shows a unilocular cyst filled with keratin and desquamated cellular material and a fibrous cyst wall lined

This article is available from: <http://www.clinicalimagingscience.org/content/1/1/4>

Copyright: © 2011 Loberant N. This is an open-access article distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited.

This article may be cited as:

Loberant N, Bhatt S, Messing E, Dogra VS. Bilateral Testicular Epidermoid Cysts. J Clin Imaging Sci 2011;1:4
Available FREE in open access from: <http://www.clinicalimagingscience.org/text.asp? 2011/1/4/73502>

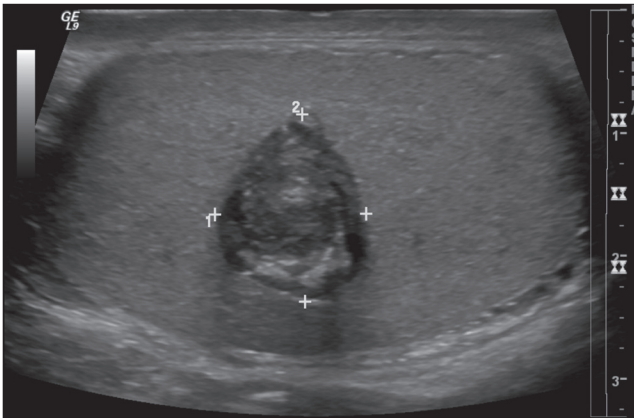


Figure 1: High-frequency transducer gray-scale sonogram of the right testis demonstrates an intratesticular mass measuring 1.2 cm × 1.5 cm. The mass predominantly appears solid and has minimal lamellation. This mass did not demonstrate any vascularity within it.

by squamous epithelial cells, with no evidence of testicular intratubular neoplasia.^[1]

Complete resection of the tumor is curative with no evidence of recurrence or metastasis. Given the striking difference between TEC and germ cell tumors, the necessity for accurate diagnosis becomes clear because testicular malignancy necessitates orchiectomy and TEC can potentially be managed more conservatively. Previously, epidermoid tumors had consistently been managed with orchiectomy because of uncertainties concerning diagnosis.^[4] In recent years, reports of successful treatment with testis-sparing surgery have become more numerous. Either enucleation or wedge resection has been performed, with concomitant use of intraoperative frozen section histologic analysis, to rule out occult malignancy within the tumor and in the adjacent testicular parenchyma (testicular intratubular neoplasia or carcinoma *in situ*).^[2,5-7]

A number of reviews and case reports have been published during the past 10 years that describe the imaging of TEC.^[8-12]

Imaging of TEC, as in scrotal lesions in general, begins with sonography. Usual features include a sharply defined unilocular cystic mass 1–3 cm in diameter with a hyperechoic rim. Within the lesion, there is often observed a unique "onion ring" appearance, first described by Malvica,^[13] of alternating hypoechogenicity and hyperechogenicity representing layers of compacted keratin and desquamated squamous cells. Alternatively, many cases of TEC show a heterogeneous echotexture with a target or bull's eye pattern consisting of a hyperechoic capsule, hypoechoic content of fat and water, and a hyperechoic central focus representing keratin debris.^[8] It is uncommon to see calcifications.^[4] Color Doppler examination consistently reveals TEC to be avascular,^[3,12] unlike most testicular malignancies.

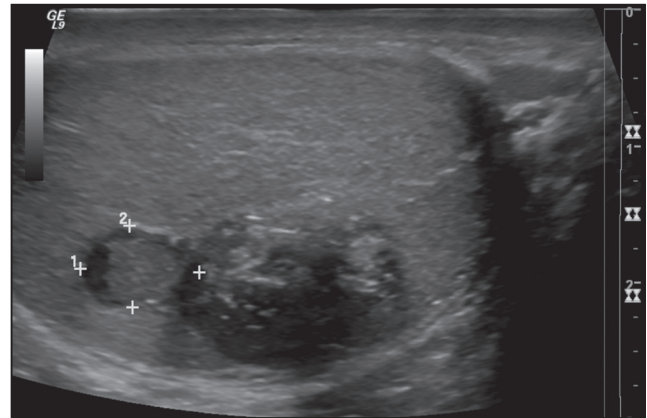


Figure 2: High-frequency transducer gray-scale sonogram of the left testis demonstrates an intratesticular mass. This mass has variable echotexture with areas of increased echogenicity anteriorly. There is an additional small lesion (within calipers) connected with this main mass.

Magnetic resonance (MR) imaging has also been described in TEC,^[3,4] and a similar finding of laminated high- and low-signal intensity corresponds to the pathologic finding of multiple layers of keratin debris. However, rather than this "classic" appearance, as on ultrasound, a cystic mass with irregular capsule may be observed.^[3]

Unfortunately, there may be overlap in the sonographic appearance of TEC, usually lined exclusively by squamous epithelium, and teratoma, in which there is prominent squamous epithelium (ectodermal origin) and cylindrical epithelium (endodermal origin) lining the cyst.^[8] The latter histologic finding removes the tumor from the clearly benign category and necessitates aggressive treatment. In addition, TEC occasionally appears atypical, with a multilocular configuration, without the onion ring or target appearance.^[8] In a patient with TEC, additional lesions must be sought assiduously because of the possibility of synchronous teratoma.^[14] However, the situation is further complicated by the existence of mature teratoma in pre-pubertal boys, which, like TEC, is derived from benign germ cells and, like TEC, is a benign lesion.^[15] In this lesion, there is no evidence of intratubular germ cell neoplasia (IGCN), whereas in 90% of the post-pubertal teratomas IGCN is found histologically. There is strong evidence that ovarian and pre-pubertal testicular teratomas are derived from benign germ cells, a pathogenesis that likely also applies to rare dermoid cysts and uncommon epidermoid cysts of the testis.

Concern that the testicular lesion represents malignancy may lead to a search for metastases using computed tomography, MR or positron emission tomography; these examinations will be negative in TEC. In addition, tumor markers (β -HCG, AFP) are negative in patients with TEC.^[5,10,11]

When the pre-operative imaging findings suggest TEC

and intraoperative frozen sections confirm the absence of malignancy, conservative, testicular-sparing surgery may be used safely.

The correct diagnosis and management becomes even more imperative in bilateral epidermoid cysts. Bilateral TEC is a very rare condition, with only a few reports in the literature.^[16-19] In all, pre-operative imaging suggested the correct diagnosis and testis-preserving surgery, using enucleation or wedge resection, was performed for most of the involved testes. Bilateral TEC presents a unique challenge in treatment as the previously accepted protocol would entail bilateral orchiectomy. However, with normal tumor markers, benign imaging appearance and benign intraoperative histology, testis-sparing surgery becomes possible.

Our patient had bilateral avascular intratesticular masses and negative tumor markers. The mass in the left testis was palpable and multifocal; the mass in the right testis was unifocal and not palpable. The ultrasound image did not display the classic onion ring pattern but did show some lamellation. The main pre-operative differential considerations were congenital bilateral adrenal rests, bilateral epidermoid cysts, bilateral germ cell tumors or a combination of lesions. It was decided to perform open biopsy of the left testicular mass as it was palpable. During surgery, the mass appeared to be a keratin-filled epidermoid cyst. Frozen section histology was consistent with keratin-filled TEC. Final pathology of the excised testicular mass predominantly showed a keratin-filled squamous cyst. A focal cystic area was identified in the vicinity of, but apparently not in contact with, the squamous cyst. This cystic area showed the presence of elements including ciliated glandular epithelium, intestinal epithelium, and seromucinous glands. These findings were consistent with a mature teratoma [Figure 3]. The spermatogenesis appeared unremarkable within the seminiferous tubules. No evidence of intratubular germ cell neoplasia was identified. The biologic behavior of teratoma in this patient without the presence of IGCN could not be predicted with certainty. But, because of the lack of findings typically seen in post-pubertal teratomas with respect to the nature of the adjacent testicular parenchyma, and the absence of IGCN, the lesion was thought to represent a pediatric form of teratoma. This could have been present in the patient for a long period of time, developing in the pre-pubertal era but not presenting until the post-pubertal period. The specimen was also sent for 12 p amplification by a fluorescence *in situ* hybridization (FISH) study. An amplification of 12 p was not observed in the tissue, again favoring a benign teratoma.

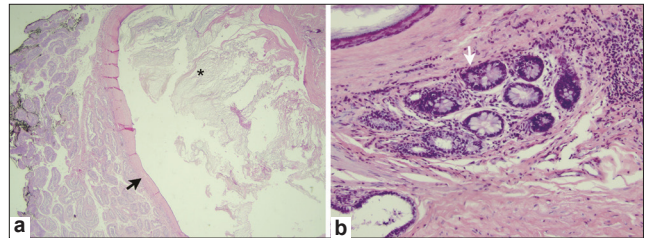


Figure 3: (a) Histopathology at 40× with hematoxylin and eosin stain demonstrates an intratesticular epidermoid cyst lined with squamous epithelium and keratin debris in the center (arrow). (b) Additional section through the testis with glandular formation (arrow) and smooth muscle cells. This is suggestive of teratomatous elements.

Thus, our conclusion was that the bilateral masses in this 17-year-old represented congenital epidermoid cysts with atypical elements rather than post-pubertal germ cell tumors. The presence of teratomatous elements in the absence of germ cell intratubular neoplasia is considered benign, the decision was made to treat conservatively and follow the patient closely. During follow-up after the initial treatment, the patient has recovered uneventfully and the ultrasound picture has been stable.

REFERENCES

1. Price EB Jr. Epidermoid cysts of the testis: a clinical and pathologic analysis of 69 cases from the testicular tumor registry. *J Urol* 1969;102:708-13.
2. Dieckmann KP, Loy V. Epidermoid cyst of the testis: A review of clinical and histogenetic considerations. *Br J Urol* 1994;73:436-41.
3. Langer JE, Ramchandani P, Siegelman ES, Banner MP. Epidermoid cysts of the testicle: sonographic and MR imaging features. *AJR Am J Roentgenol* 1999;173:1295-9.
4. Cho JH, Chang JC, Park BH, Lee JG, Son CH. Sonographic and MR imaging findings of testicular epidermoid cysts. *AJR Am J Roentgenol* 2002;178:743-8.
5. Li WM, Su CM, Kang WY, Wu WJ, Chou YH, Huang CH. Conservative surgery for epidermoid cyst of the testis: A case report and recent literature review. *Journal of Taiwan Urological Association*. 2008;19:242-4.
6. Shukla AR, Woodard C, Carr MC, Huff DS, Canning DA, Zderic SA, et al. Experience with testis sparing surgery for testicular teratoma. *J Urol* 2004;171:161-3.
7. Ross JH, Kay R, Elder J. Testis sparing surgery for pediatric epidermoid cysts of the testis. *J Urol* 1993;149:353-6.
8. Maizlin ZV, Belenky A, Baniel J, Gottlieb P, Sandbank J, Strauss S. Epidermoid cyst and teratoma of the testis: Sonographic and histologic similarities. *J Ultrasound Med* 2005;24:1403-9.
9. Strauss S, Gottlieb P, Kessler A, Graif M, Heyman Z, Manor H. Non-neoplastic intratesticular lesions mimicking tumour on ultrasound. *Eur Radiol* 2000;10:1628-35.
10. Loya AG, Said JW, Grant EG. Epidermoid cyst of the testis: Radiologic-pathologic correlation. *Radiographics* 2004;24:S243-6.
11. Dogra VS, Gottlieb RH, Rubens DJ, Oka M, Di Sant Agnese AP. Testicular epidermoid cysts: sonographic features with histopathologic correlation. *J Clin Ultrasound* 2001;29:192-6.
12. Dogra VS, Gottlieb RH, Rubens DJ, Liao L. Benign intratesticular cystic lesions: US features. *Radiographics* 2001;21:S273-81.
13. Malvica RP. Epidermoid cyst of the testicle: an unusual

- sonographic finding. *AJR Am J Roentgenol* 1993;160:1047-8.
14. Huyghe E, Mazerolles C, Moran C, Khedis M, Khoury E, Nohra J, et al. Synchronous epidermoid cyst and mature teratoma of the testis: An unusual association. *Urol Int* 2007;78:364-6.
 15. Ulbright TM. Germ cell tumors of the gonads: a selective review emphasizing problems in differential diagnosis, newly appreciated, and controversial issues. *Mod Pathol* 2005;18: S61-79.
 16. Shenoy V, Triest WE, el-Bash O. Bilateral epidermoid cysts of the testis: report of a case with preservation of 1 testis. *J Urol* 1995;153:1933-4.
 17. Davi RC, Braslis KG, Perez JL, Soloway MS. Bilateral epidermoid cysts of the testis. *Eur Urol* 1996;29:122-4.
 18. Sloan J, Beck S, Bihrlle R, Foster R. Bilateral testicular epidermoid cysts managed by partial orchiectomy. *J Urol* 2002;167:255-6.
 19. Cittadini G, Gauglio C, Pretolesi F, Santacroce E, Derchi LE. Bilateral epidermoid cysts of the testis: sonographic and MRI findings. *J Clin Ultrasound* 2004;32:370-2.

Source of Support: Nil, **Conflict of Interest:** None declared.