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Conservative Surgical Management of Bilateral Epidermoid Cysts of the Testis: a Case Report and Review of Literature

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Introduction

Epidermoid cysts of the testis are benign tumor-like intratesticular masses. They are rare, representing less than 1% of all testicular tumors, and approximately 300 cases have been reported in the literature. Treatment of epidermoid cysts is controversial with the traditional treatment being radical orchidectomy. Awareness of this entity and the criteria for differential diagnosis will enhance the possibility of testicular preservation, averting unnecessary orchidectomy, especially for bilateral lesions. We report a case of bilateral epidermoid cysts in a 19-year-old man in whom the testes were preserved with conservative surgical management.

Case Report

A 19-year-old Bahraini man presented with bilateral testicular pain of 5 months' duration. Three years earlier, he had undergone right scrotal exploration and evacuation of a hematocele following blunt trauma to the scrotum. Physical examination revealed 2 nontender masses (2 × 2 cm) in the midportion of both testes bilaterally. They were separate from the scrotal wall and epididymides. There was no clinically detectable hydrocele, gynecomastia, or palpable abdominal or inguinal mass. High-resolution testicular ultrasonography showed a 1.6 × 1.5 cm well-defined, spherical hypoechoic bilateral lesions with peripheral hyperechogenicity and normal surrounding parenchyma. Serum levels of tumor marker (β-human chorionic gonadotropin and α-fetoprotein) were in the reference ranges. The patient underwent bilateral inguinal exploration. A soft clamp was placed on the spermatic cord, prior to delivery of the testis from the scrotum. The testes were bisected, and well-circumscribed cysts were identified. The lesions were completely enucleated. On incision of the cysts, yellow-white, putty-like material was extruded, revealing smooth cystic lesions (Figure 1). The testes were replaced in the scrotum after tunical closure. Frozen section histopathologic examination of both lesions confirmed the diagnosis of an epidermoid cyst. Histologic examination of paraffin sections validated this (Figure 2). The patient's postoperative course was uneventful, and he was well on follow-ups at 3, 6, and 12 months. Results of analysis of his semen are normal 1 year after the surgery, and no recurrence has been detected clinically or by ultrasonography.

FIG. 1. Macroscopic appearance of the epidermoid cyst filled with pale pasty material

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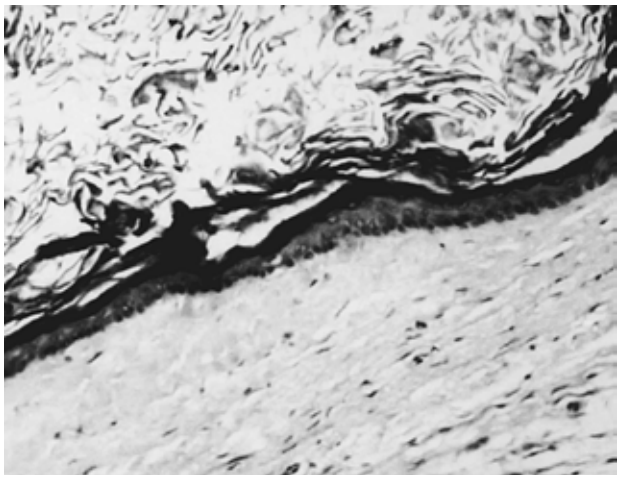

Fig. 2. Pathologic examination shows epidermoid lining with distinct granular layers and keratin flakes filling the lumen of the cyst (hematoxylin-eosin, × 100).

Discussion

Epidermoid cysts of the testis are uncommon, accounting for 1.5% to 2% of testicular tumors.[2,3] Since their first report in 1942 by Dockerty and Priestly, more that 300 cases have been reported in the literature.[1] To our knowledge, this case represents the fourth reported case of bilateral epidermoid cyst in an adult patient and the third reported case with bilateral conservative management.[4,5]

The histogenesis of epidermoid cysts has not been completely elucidated; however, the prevailing hypothesis is that of germ-cell origin, developing along the line of epidermoid differentiation as a monodermal expression of a teratoma.[2,6] Other theories have been postulated, such as squamous metaplasia of the seminiferous epithelium, metaplasia of the rete testis, and inclusion of epidermoid cysts.[2,6] Its relationship with other germ cell tumors is supported by the age and racial distribution of patients affected, and by its reported occurrence in the ovary and cryptorchid testes.[2]

Diagnosis of a lesion as a simple benign epidermoid cyst of the testis is confirmed pathologically when the following 5 criteria, set by Prince in 1969, are met: the lesion is a cyst located within the parenchyma of the testis; the lumen is filled by keratinized debris; the wall of the cyst is composed of fibrous tissue with complete or incomplete lining of squamous epithelium; no teratomatous elements or dermal adnexal elements are present either within the cyst wall or elsewhere within the parenchyma of the testis; and no scar is present in the remaining testicular parenchyma.[2] Strict adherence to these criteria clearly separates this benign entity from the more complex bidermal and tridermal testicular teratomas, all of which have malignant potential.[2] Pathologically, an epidermoid cyst is typically a round or oval encapsulated lesion. The lumen contains squamous epithelium and keratin debris that may vary from complex fluid to a thick, pasty consistency.[6] The wall is composed of fibrous tissue that may be calcified.[6]

Epidermoid cysts of the testis usually present as incidental findings of a painless nodule during self-examination or routine physical examination.[1] Most epidermoid cysts of the testis (86%) occur in young men in the second to fourth decades of life, but neither the young nor the aged are spared.[2] Clinically, epidermoid cyst cannot be differentiated from other testicular tumors. They usually measure 2 to 3 cm on average, with a slight predominance to the right testis, similar to germ cell tumor.[1] Bilateral occurrence is rare. Clinically, an epidermoid cyst appears as a circumscribed, firm, smooth, and nontender intratesticular mass.[1] They apparently have no predilection for central-hilar parts rather than polar regions of the testis.[2] Tumor markers, such as serum β-human chorionic gonadotropin and α-fetoprotein, are negative.[6]

Scrotal ultrasonography is a common diagnostic procedure for evaluating palpable scrotal masses to distinguish intratesticular from extratesticular lesions, and to exclude contralateral lesions. The ultrasonographic characteristics of epidermoid cysts include a well-circumscribed hypoechoic mass with a hyperechogenic rim.[7] The ongoing deposition of keratinized material inside the cyst creates an echogenic whorl forming an onion-skin typical appearance.[1,7] The wall may calcify, and it may even develop a bony shell.[6] Absence of flow on color Doppler ultrasonography is consistent with the avascular nature of these lesions. Several authors have found typical patterns that are highly suggestive of malignancy rather than benign conditions. The presence of a hypoechoic mass and bright echoic foci, as well as heterogeneous parenchymal echo patterns, all have been found to be suspicious for tumors.[8] On magnetic resonance imaging, epidermoid cysts show concentric rings of alternating high-signal and low-signal intensity corresponding to

the pathologic finding of multiple layers of keratin.[1,6] There is no contrast enhancement.[6]

Testicular lesions have traditionally been treated by radical orchidectomy when any mass was detected, but up to a third of these operations may be unnecessary.[8] Treatment of epidermoid cysts remains controversial. It may be that the controversy regarding the histogenesis of epidermoid cysts has had a direct effect on the surgical approaches. Nevertheless, radical orchidectomy has been the gold standard, although is now less frequently used.[1,3-6,9,10] Indeed, more conservative approaches based on testis-sparing surgery are gaining more acceptance. A most recent literature review shows that almost all of reports published in the last decade on this subject invariably emphasize the safety and importance of conservative treatment of epidermoid cysts in all age groups.[1] The rationale of a less aggressive approach is that no local recurrence, distant metastases, or deaths have been reported in patients treated conservatively, from follow-up information available for up to 37 years.[2,3] Many factors have led to this shift in the surgical approach. The typical ultrasonographic features of epidermoid cyst have facilitated a more accurate preoperative diagnosis allowing preoperative planning of testicular preservation.[7] The high accuracy of intraoperative frozen section is another factor that has promoted safe testicular preservation.[1] Moreover, the macroscopic appearance of an epidermoid cyst at the time of testicular exploration is typical, with a light yellowish-tan well-encapsulated tumor that contains keratin plug and shells out easily from the testis.[9,10] If the ultrasonographic characteristics before surgery, age, physical examination, and tumor markers are in concordance with a strong possibility that a given mass is benign, a limited inguinal exploration and subsequent frozen-section analysis can be used to reduce the number of unnecessary orchidectomies.[8] Considering the psychological implications, preservation of a testicle in a young man is crucial. Furthermore, most patients are of reproductive age, and preservation of fertility is a major issue in testicular surgery. However, it should be emphasized that all solid intratesticular lesions must be presumed as being malignant. The principles of cancer surgery must be followed, the testis should be isolated with nontraumatic clump, and the wound protected from potential spillage of tumor cells. The tunica albuginea should be opened over the mass, and the tumor must be excised in-toto and submitted for frozen section examination.

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