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CASE REPORT

# Testis Sparing Surgery for Bilateral Epidermoid Cyst Associated with Testicular Microlithiasis

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ABSTRACT

Testicular epidermoid cyst is a benign epithelial tumor rarely seen in children. Recurrence or distant metastasis after excision is not reported. Testis sparing surgery is important when there is bilateral involvement. A 13-year-old boy with bilateral testicular epidermoid cyst associated with microlithiasis treated with testis sparing surgery is reported with special emphasis on differential diagnosis and treatment.

Key words: Testis, surgery, epidermoid cyst, testicular microlithiasis

## INTRODUCTION

Epidermoid cysts of testis are rare benign tumoral lesions accounting for 3-14% of all pediatric testicular tumors. They are mostly seen on third and fourth decades of life and rarely seen in adolescent boys [1,2]. Right side is more commonly involved than left and bilateral occurrence is exceedingly rare. Epidermoid cyst of testis typically exhibits a benign clinical course so testis-sparing surgery is preferred rather than orchiectomy. However, the association between testicular microlithiasis and germ cell tumors may lead misinterpretation and unnecessary orchiectomy in a child with epidermoid cyst and coexisting testicular microlithiasis. Thus, differentiation of a testicular epidermoid cyst and a testicular germ cell tumor is a diagnostic challenge in the presence of testicular microlithiasis. Testicular microlithiasis is a condition characterized by calcium deposition in the lumina of seminiferous tubules [3]. It is being more recognized with the wide use of scrotal ultrasonographic examination. There are ongoing investigations about the etiology of microlithiasis and the association between microlithiasis and testicular tumors. Herein, the authors present a case with bilateral testicular epidermoid cyst and bilateral testicular microlithiasis treated with enucleation of epidermoid cysts.

## CASE PRESENTATION

A 13-year-old boy presented to his pediatrician with the complaint of obesity and during the physical examination, the physician palpated bilateral nontender solid scrotal masses. Scrotal ultrasound revealed 20x15 mm in diameter hyperechoic heterogeneous solid- cystic mass in right testis and 10x11 mm in diameter mass with the same characteristics as on the right side located in the left testis. These both lesions were intraparenchymal and the remaining testicular tissue contained microlithiasis (Figures 1 and 2). Epididymal structures were normal. Tumor markers including alphafetoprotein and human chorionic gonadotropin levels were normal. Patient was operated with bilateral inguinal approach. Spillage and seeding was avoided by occlusion of spermatic

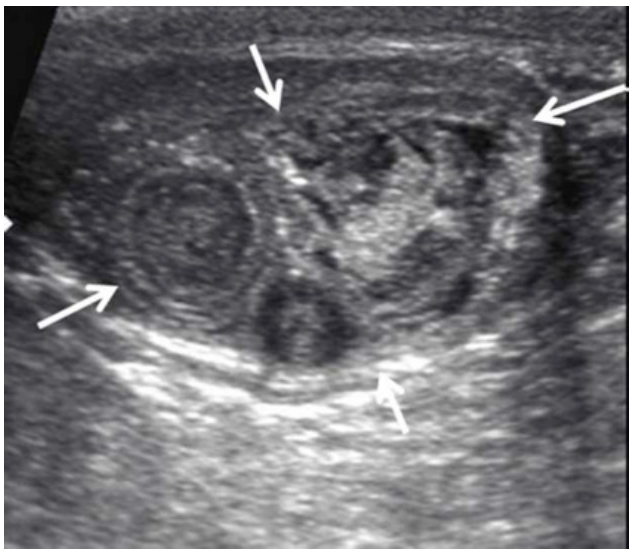

Figure 1. Epidermoid cyst containing alternating hyperechoic and hypoechoic internal rings with an onion skin appearance.

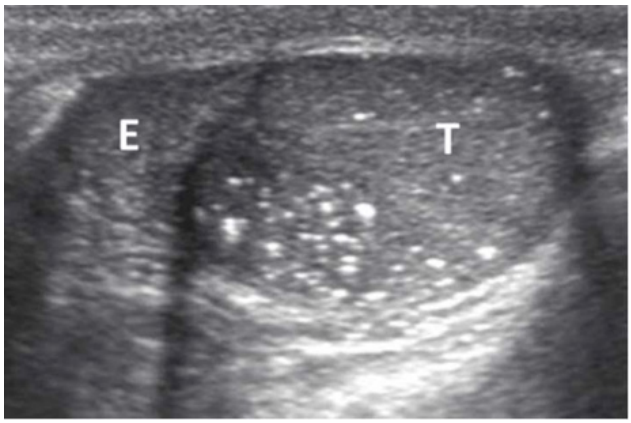


Figure 2. Multiple echogenic foci demonstrating testicular microlithiasis.

cord by the help of a Penrose drain, which is used as a tourniquet, and isolation of surgical field by sterile surgical drapes. Evaluation of frozen sections of the excised cystic lesions revealed epidermoid cyst. Thus bilateral testis sparing surgery with enucleation of the masses was performed. The final pathologic report confirmed the diagnosis (Figure 3). Patient had an uneventful postoperative period with no recurrence during 18 months.

## DISCUSSION

Testicular epidermoid cyst is a rare benign epithelial intratesticular tumor and it accounts for 3-14% of all testicular tumors in children [1]. Docherty and Priestly described the entity in 1942. The pathogenesis is uncertain but it is hypothesized that

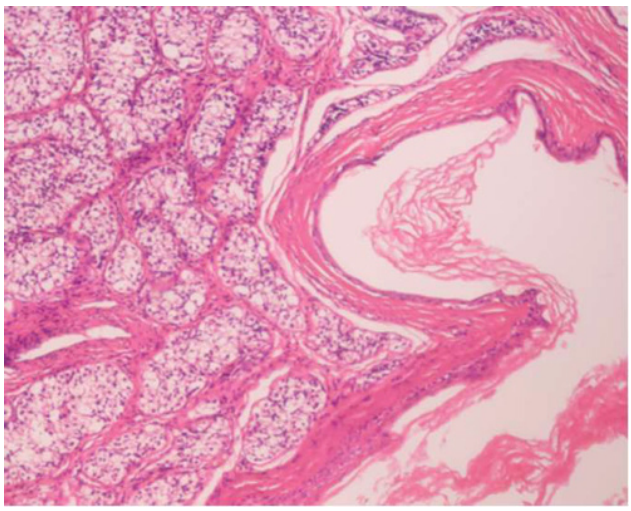

Figure 3. Fibrous wall separating the cyst from the surrounding semiferous tubules.

epidermoid cysts originate from a germ cell developing along epidermal differentiation and representing a monodermal teratoma. It lacks endodermal and mesodermal components or skin appendages differentiating it from classic teratoma and dermoid cyst [1,2]. Unfortunately, the clinical manifestations of epidermoid cysts are identical to those of germ cell tumors typically presenting as nontender palpable intratesticular mass. The patients may be asymptomatic and epidermoid cyst may be detected during routine physical examination. Usually the only presenting symptom is painless scrotal swelling. On physical examination usually a nontender round smooth peripherally located intratesticular mass is palpated. Ultrasonographic imaging shows a well-demarcated intraparenchymal mass with variable echogenicity. A target appearance or onion ring configuration can be detected depending on the echogenicity pattern of the lesion. Universally, the lesion lacks vascular supply and this finding reduces the risk of malignancy in the preoperative assessment [4-7]. The tumor markers such as alphafetoprotein and human chorionic gonadotropin levels are normal [2]. In the past, radical orchiectomy was the standard treatment plan but nowadays with the advance of ultrasound imaging, testis- sparing surgery is gaining more popularity especially for children [8-10]. It is well known that if the lesion is proven to be a simple epidermoid cyst on pathological examination, clinical course is totally benign without concern for local recurrence or distant metastasis. If the preoperative evaluation suggests epidermoid cyst strongly in the setting

of benign ultrasonographic appearance and negative tumor markers, inguinal approach and perioperative frozen section histologic analysis of the enucleated lesion may be the reasonable choice rather than radical orchiectomy. There are certain histologic criteria stated by Price in order to diagnose testicular epidermoid cysts which are; intraparenchymal localization, lumen containing keratinized material, fibrous wall, no teratomatous elements or skin appendages, no scar in the remaining testiscular tissue [1,2].Testicular microlithiasis is a condition characterized by calcium deposition in the lumina of seminiferous tubules, which form from degenerated cells. The etiology of this lesion is unknown. Drut et al suggested testicular microlithiasis is related with sertoli cell dysfunction and abnormal gonadal embryogenesis during the early stages of testicular development [11]. These lesions are seen on ultrasound as multiple speckled echogenic foci within the testis, which can be focal or diffuse. The widespread use of ultrasound imaging increased the frequency of detection of testicular microlithiasis. Incidence in pediatric population ranges from 1.1 to 4.2%. Association of testicular microlithiasis and testicular tumors is studied and they do not yield relevance for adults and this is also the case for children. But there are also case reports of testicular microlithiasis associated with malignant tumors among adolescents [12]. Nonetheless, authors advise a regular follow up strategy with physical examination and ultrasound imaging and instructing children for self-examination. Long term prospective studies are needed to produce evidence based algorithm. This topic is beyond the scope of this report. Hughes et al presented a case report of a 12-year-old patient with testicular microlithiasis and epidermoid cyst [10]. We wonder if the epidermoid cysts become in order to restrict microliths or otherwise microliths evolve as an inflammatory response to epidermoid cysts. Unfortunately, our patient does not have a previous scrotal ultrasound so we do not know whether these lesions are synchronous or metachronous.Testicular epidermoid cyst is a rare benign epithelial tumor with no recurrence or distant metastasis after excision. When ultrasonographic imaging findings suggest that the intratesticular mass is likely to be an epidermoid cyst the patient should be offered testis sparing surgery rather than orchiectomy. Testis sparing approach gains more importance when the lesion is bilateral. However, the possible association between testicular microlithiasis and malignant tumors may lead a diagnostic and therapeutic challenge in a child with testicular cystic lesion and coexisting testicular microlithiasis. Thus, differentiation of a testicular epidermoid cyst and a testicular germ cell tumor is essential. If the preoperative evaluation by ultrasonographic examination and tumor markers suggests epidermoid cyst, inguinal approach and perioperative frozen section histologic analysis of the enucleated lesion may provide testis-sparing surgery.

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