

Epidermoid cyst discovered concomitant with torsion of the spermatic cord: A case report

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Abstract

Testicular epidermoid cysts, accounting for 1%–2% of all testicular masses, present a diagnostic challenge often identified post-orchietomy. Conservative surgery is recommended, emphasizing the need for accurate preoperative diagnosis. A 28-year-old patient with acute left scrotal pain, initially suggestive of testicular torsion, underwent intraoperative exploration revealing extra-vaginal torsion with an incidentally discovered 2-cm intra-testicular mass. Due to suspected malignancy, a total orchietomy was performed. Pathological analysis confirmed an epidermoid cyst with normal postoperative tumor markers. The patient had a good postoperative outcome and underwent testicular prosthesis placement 3 months after the surgery. Epidermoid cysts, often identified incidentally during urgent scenarios, underscore the importance of accurate preoperative diagnosis. Conservative surgery remains the preferred approach, emphasizing the consideration of benign factors to prevent unnecessary orchietomies.

Keywords

Epidermoid cyst, testicle, spermatic cord torsion, testis sparing surgery

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Introduction

Testicular epidermoid cysts (TECs) are an uncommon benign testicular tumors, accounting for about 1%–2% of all testicular masses.¹ It is more common in the third and fourth decades of life; however, it can occur at any age.² Most of the current knowledge is derived from limited clinical series and individual case reports.³

When a diagnosis is determined through frozen-section analysis during surgery, conservative surgery is advised for TECs.⁴ However, due to the challenges in making this specific diagnosis, especially when faced with misleading presentations, they are commonly identified only after orchietomy has been performed.

We present a case involving a 28-year-old patient who presented with symptoms suggestive of testicular torsion, wherein an epidermoid cyst was incidentally discovered.

Case presentation

An unmarried 28-year-old patient with no children was admitted to the emergency room with a sudden onset of pain in the left scrotal region. The patient had no specific medical or surgical history, and no toxic habits were reported.

On physical examination, the patient was hemodynamically stable and had no fever. Pain was assessed as 10/10 on the visual analog scale.⁵ Examination of the scrotum revealed a slightly enlarged, hard, non-inflammatory left bursa. The left testicle was painful to palpation, fixed, retracted, and elevated compared with the contralateral testicle. The cremasteric reflex was absent, and Prehn's sign⁶ was negative, achieving a TWIST score of 6 (>5 = high risk).⁷ The abdomen was supple, and the hernial orifices were free. There

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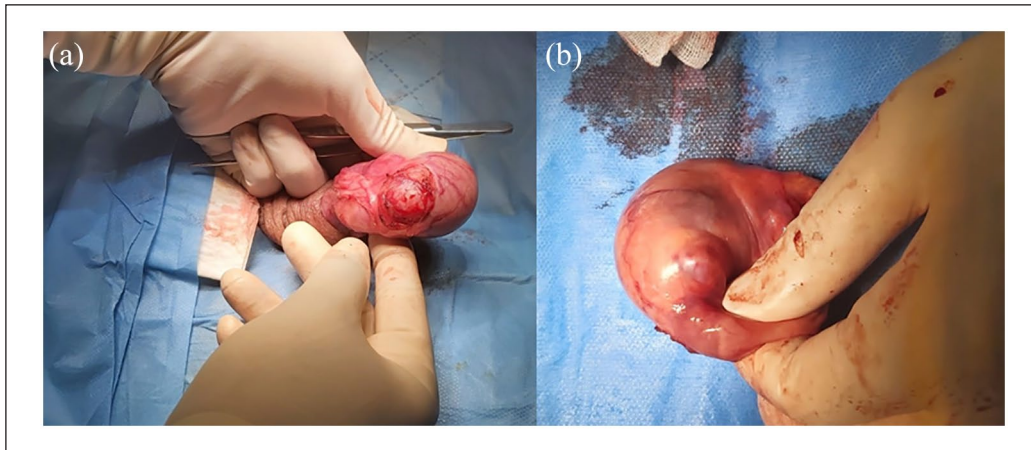


Figure 1. Inferior polar intra-testicular mass adjacent to the left testicle.

was no urethral discharge, and the rectal examination was unremarkable.

The patient reported that the pain had started abruptly 3 h before admission. It progressed continuously, radiating upwards toward the groin area on the same side, without any relief from changing position. The patient did not report any associated urinary or digestive symptoms.

Considering the clinical presentation suggestive of torsion of the spermatic cord, the decision was made to conduct a surgical exploration of the scrotum. This exploration revealed an extra-vaginal torsion of the left spermatic cord involving two counterclockwise turns of the coil, without any indications of testicular necrosis. Following the manual detorsion of the testicle and irrigation with warm saline, palpation of the testicle identified a suspicious inferior polar intra-testicular mass measuring two centimeters in length and respecting the albuginea (Figure 1). Due to the suspected malignancy of the tumor and the unavailability of an intra-operative pathological frozen section analysis, a total orchiectomy was performed, with no postoperative complications observed.

The anatomopathological analysis revealed a macroscopic appearance suggestive of a yellowish, cystic encapsulated lesion measuring $2 \times 2 \times 3$ cm, exhibiting no abnormalities in the testicular parenchyma. Microscopic examination demonstrated that the cystic formation was lined with stratified, keratinized squamous epithelium, devoid of significant cytonuclear atypia. The contents comprised eosinophilic and basophilic lamellae, supported by a fibrous membrane (Figure 2).

The anatomopathological study resulted in the final diagnosis of an epidermoid cyst of the testis, displaying no morphological signs of malignancy. Furthermore, postoperative measurements of tumor markers—alpha-fetoprotein (AFP), lactate dehydrogenase (LDH), and human chorionic gonadotropin (HCG)—were within normal ranges.

The patient had a good postoperative outcome. There had been no recurrence according to ultrasonography 3 months

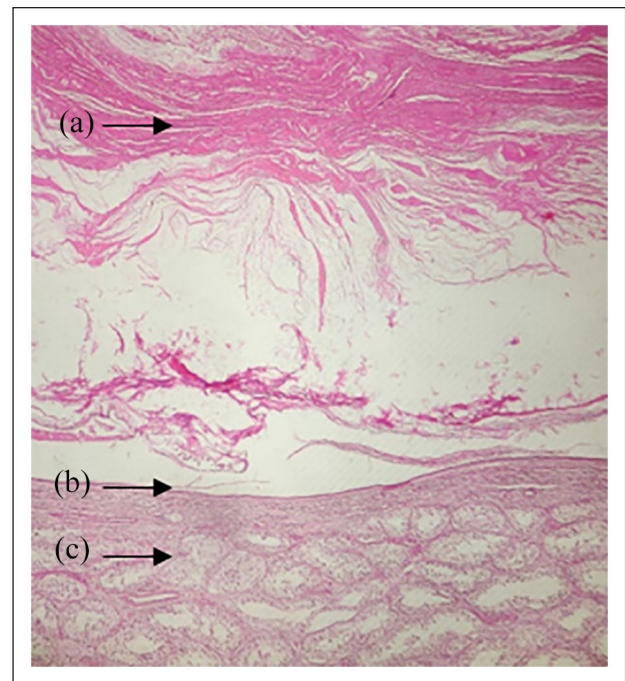


Figure 2. Pathological appearance favors an epidermoid cyst of the testis. (a) Cystic formation bordered by stratified and keratinized squamous epithelium, containing lamellae of orthokeratotic keratin. (b) Squamous lining with preserved architecture. (c) Small fibrous connective tissue resting on testicular parenchyma made up of testicular tubules without atrophy.

after surgery. He underwent testicular prosthesis placement 3 months after the surgery.

Discussion

TECs have been first reported in 1942,⁸ the histogenesis of these cysts remains unclear, with some authors suggesting they represent monodermal development of a teratoma,

whereas others opine that they arise from metaplasia of the rete testis or seminiferous epithelium.⁹

Our presented case met the aforementioned pathological criteria established by Price¹⁰ including the lesion is identified as a cyst situated within the parenchyma of the testes. The lumen is expected to contain keratin without teratomatous elements. The cyst wall should be composed of fibrous tissue, exhibiting either a complete or incomplete inner lining of squamous epithelium. Neither teratomatous elements nor adnexal structures are observed within the cyst wall or the testicular parenchyma. Occasionally, a mild lipogranulomatous inflammatory reaction with focal calcification in the wall may be present. Although the testicular parenchyma may experience atrophy, the cyst wall remains discrete and separate from the tunica albuginea.

Epidermoid cysts often present a clinical challenge due to their varied manifestations. Although most patients remain asymptomatic and incidentally discover a testicular mass during self-examination or routine physical checks, some exhibit symptoms such as scrotal pain, enlargement, or vague discomfort.⁹ However, scrotal pain does not seem to reliably differentiate between benign and malignant lesions, as evidenced by an analysis showing similar frequencies of pain across both types. Additionally, the duration of symptoms in patients with benign tumors is significantly longer than those with malignant germ cell tumors, implying a differential growth rate between the two lesion types.¹¹ Furthermore, differentiating epidermoid cysts from other testicular tumors presents a challenge due to their similar clinical presentation as a nontender, smooth, palpable mass, emphasizing the necessity for a comprehensive clinical approach to ensure an accurate diagnosis.¹ In addition, certain atypical forms have been described, notably the case of an epidermoid cyst revealed by hematospermia,¹² and two cases of benign tumors mimicking testicular torsion in triorchidism.^{13,14}

Alongside clinical evaluation, biological understanding and imaging are pivotal in surgical planning, as the choice of the appropriate technique relies heavily on considering these factors. Notably, tumor marker tests such as α -fetoprotein and HCG typically yield negative results for epidermoid cysts. Ultrasound reveals diverse characteristics,¹⁵ and these include a range of presentations such as a mass displaying a target appearance—a central hypoechoic area surrounded by an echolucent rim. Additionally, they may manifest as an echogenic mass with dense acoustic shadowing due to calcification, or present as a well-circumscribed mass with a hyperechoic rim.¹⁵ Other ultrasound patterns associated with epidermoid cysts involve a mixed pattern, displaying heterogeneous echotexture and a poorly defined contour. In addition, the “onion peel appearance,” first described in 1993 by Malvica,¹⁶ stands out as a distinctive feature in diagnosing epidermoid cysts. This pattern notably corresponds with the pathological discovery of multiple layers of keratin debris generated by the cyst’s lining. It is recognized by the presence of alternating rings of hyperechogenicities and

hypoechogenicities within the mass, offering a characteristic ultrasound signature for identifying these cysts.¹⁵

On magnetic resonance imaging (MRI), the typical appearance is the “bull’s eye.” The wall and the center of the cyst are in hypo-signal in T1 and T2 weighting. Between these two zones, there is, in T1 and T2 weighting, a hyper-signal that corresponds to isolated and degenerated epithelial cells.¹⁷

Currently, the recommended treatment for benign testicular tumors involves conservative surgery, either through enucleation or partial orchiectomy, following a conclusive intra-operative pathological frozen section analysis. This approach aims to preserve fertility and minimize the psychological impact of orchiectomy. It is noteworthy that several studies have indicated no subsequent recurrence.¹⁸ Nevertheless, orchiectomy remains a pertinent consideration in managing testicular tumors due to the prevalence of malignant forms. Hence, it is crucial to identify factors supporting benignity to avoid unnecessary orchiectomy procedures. Additionally, apart from suggestive radiological criteria (ultrasound and MRI), normal serum tumor markers (AFP, HCG, and LDH), and a conclusive negative frozen section examination, studies¹⁹ suggest other criteria. These may include lesions with a tumor volume less than 2.8 cm³, a history of hormone disorders or infertility, and a prolonged duration of symptoms.¹⁹ Furthermore, Pattamapaspong et al.⁹ propose that performing two biopsies of surrounding testicular parenchyma, revealing no associated malignancy in patients with tumors smaller than 3 cm, can aid in diagnosis.

Our reported case presents some limitations, the incidental discovery of the mass during a testicular torsion did not allow for the establishment of an appropriate preoperative approach. Indeed, a preoperative ultrasound scan could have diagnosed the testicular tumor. However, the reported case had a TWIST score > 5, indicating a high risk of testicular torsion.⁷ This is why we opted for surgical exploration from the outset, in addition to some socio-economic reasons. Additionally, the unavailability of intra-operative pathological frozen section analysis in the emergency setting prevented the identification of presumptive benign factors. Despite the viable appearance of the testicle after detorsion, the decision to perform a total orchiectomy was made intra-operatively due to the high probability of malignant forms in testicular tumors. However, we believe that biopsies should always be performed to confirm malignancy when deciding on orchiectomy (wherever possible).

Conclusion

This case highlights the diagnostic and therapeutic challenge posed by its atypical presentation. Although epidermoid cysts are among the rare benign testicular tumors, the search for potential benign factors remains crucial in order to avoid unnecessary orchiectomies and to favor conservative surgical approaches.

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Author contributions

G. E. O. and H. R. took part in taking care of the patient, taking photos, researching the bibliography, and writing the article. O. L. participated in the bibliographic research. M. R. participated in writing case presentation. M. K. and B. E. K. contributed to the interpretation of the anatomopathological elements. A. H. participated in the design and approved the final manuscript. All the authors have approved the final version of the manuscript.

Availability of data and material

On request, email the corresponding author.

Code availability

Not applicable.

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Ethical approval

Our institution does not require ethical approval for reporting individual cases or case series.

Informed consent

Written informed consent was obtained from the patient for his anonymized information to be published in this article.

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