

Atypical Testicular Torsion Due to an Epididymal Cyst in a Young Adult: Case Report

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Background: Acute scrotal pain is a urological emergency. Testicular torsion (TT) is a surgical emergency that compromises rapidly testicular viability. There is approximately a 4- to 8-hour window from the onset of torsion symptoms until surgical intervention is required to save the affected testis. Delays in care may necessitate orchiectomy, which has been associated with reduced fertility. It can occur at any age, with two peaks of frequency: during the first years of life and at puberty. The causes of testicular torsion are multiple and are related to a defect in the fixation system of the testicle.

Case Presentation: Here we report an unusual situation of testicular torsion induced by a large epididymal cyst in a young adult. The cyst was excised as well as bilateral testicular fixation and the patient was discharged on the third day. Clinical examinations and Doppler ultrasound at the sixth and twelfth postoperative weeks were both normal.

Conclusion: This observation leads us to consider epididymal cysts as a risk factor for TT.

Keywords: atypical, testicular, torsion, epididymal cyst, young adult

Introduction

Described for the first time by Hunter in 1810, testicular torsion or torsion of the spermatic cord is a formidable surgical emergency. Testicular torsion (TT) is rotation of the testis around its pedicle with disruption of blood supply and venous return, compromising viability.¹

Spermatic cord torsion is responsible for acute ischemia of the testis, leading to loss of vitality if vascular circulation is not urgently restored. It can cause significant damage to the contralateral testis through ischemia-reperfusion lesions, affecting the patient's fertility.²

Functionally, exocrine activity is highly sensitive to ischemia, and spermatogenesis is rapidly compromised by early, irreversible damage to the seminiferous tubules, usually beyond six hours.

Conversely, endocrine activity can be maintained after prolonged ischemia.³ In adults, TT are less frequent and account for about 39% of cases.⁴ T.T. can occur at any age, but two peaks of incidence are recognized: the first year of life and the pubertal period.^{2,3,5} The annual incidence of TT is approximately from 3.5 to 25 per 100,000 person-years for boys aged younger than 18 years. Two peaks of the age-specific incidence are in young children and adolescents; the incidence decreases after age 18 years.⁶⁻⁸

In published pediatric data, the incidence of torsion has been reported to be 4.5 per 100,000 males aged 1 year to 25 years annually in the USA.^{6,9}

Treatment is surgical and should ideally be performed before six hours.³ However, the six-hour delay is a statistical concept and not a safety time.¹⁰

The typical clinical presentation consists of unilateral testicular pain of acute onset in a non-febrile context. Physical examination usually reveals a large painful bursa with elevated and horizontalized testis (positive Gouverneur's sign).

Elevation of the testis does not relieve the pain (negative Prehn's sign). The epididymis is tender, while palpation of the cord sometimes reveals spires.^{1,4,11}

Scrotal ultrasound combined with Doppler will assess both vascular flow and the presence of spires in the spermatic cord; however, a normal ultrasound scan does not formally exclude the diagnosis of testicular torsion and should not delay surgical treatment.^{3,12}

The causes of testicular torsion are varied and are related to a defect in testicular fixation, such as an excessively long spermatic cord, absence of the testicular gubernaculum or mesorchium (the testicle is attached only at its upper pole, resulting in a "bell-shaped" configuration), or a defective epididymo-testicular junction.

It may also be due to an imbalance between the volume of the testis and its attachment structures, a condition known as testicular inversion.¹²

In rare cases, torsion may result from a defective attachment of the tunica vaginalis to the other sheaths of the bursa.¹¹

The possibility that an epididymal cyst directly promotes testicular torsion is plausible but extremely rare and poorly documented in the medical literature. We believe that a large epididymal cyst can: Alter the mobility of the testicle in the scrotal sac; Weigh down the upper pole of the testicle or change its center of gravity; Create traction or an imbalance of the spermatic cord. This can favor a rotation of the testicle around the spermatic cord, especially if other factors are present. There are no or very few published cases where the presence of an epididymal cyst is implicated as a proven cause of testicular torsion. In most cases, however, no underlying cause is found; these are known as idiopathic torsions. They may include unusual causes such as epididymal cysts, as described in this case report.

Case Report

Patient History

A 35-year-old male patient presented to the emergency department with sudden onset of pain and swelling of the right scrotal bursa for six hours.

The pain was not relieved by oral painkillers (Paracetamol and Diclofenac) taken at home, and was accompanied by episodes of vomiting.

The patient reported a gradual growth of a mass in the right bursa over the past few months, which gave him the impression of having two right testicles. He also reported several episodes of low intensity pain in the same bursa, each of them lasting approximately ten minutes.

The scrotal ultrasound performed at that time confirmed a large epididymal cyst at the upper pole of the epididymo-testicular junction (Figure 1).

Clinical Findings

On clinical examination, the right bursa was swollen; the testicle was slightly elevated and very painful. We palpated a rounded paratesticular mass of liquid consistency.

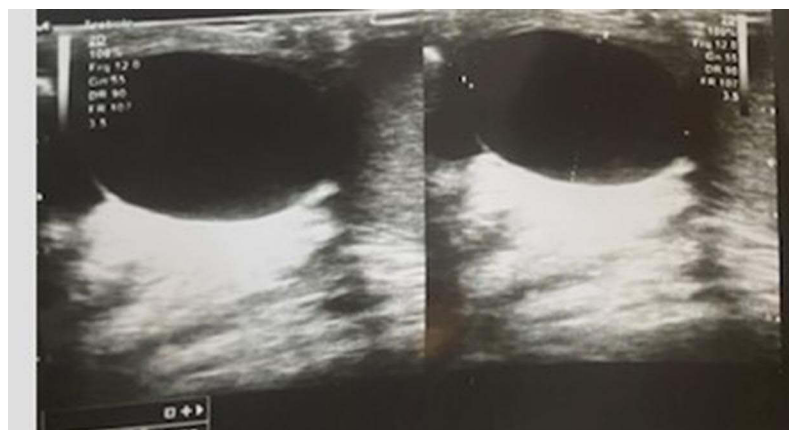


Figure 1 Large epididymal cyst at the upper pole of the epididymo-testicular junction.

The search of spires in the spermatic cord was impaired by the pain and the paratesticular liquid mass.

Diagnostic Approach

Scrotal ultrasound clearly identified a large epididymal cyst. However, Doppler imaging of the spermatic cord was significantly hampered by this large cyst.

Therapeutic Management and Follow-Up

Surgical exploration was promptly performed, with the presumptive diagnosis of a large and painful epididymal cyst. Intraoperatively, we noted three tight clockwise oriented twists in the cord. The testicle was enlarged, purplish and surmounted by a large cephalic epididymal cyst (Figure 2).

After manual untwisting, the testis recovered its normal color and arterial pulsations in the spermatic cord were readily evidenced (Figure 3).

The traditional risk factors for testicular torsion were not identified.

The cyst was excised (Figure 4), as well as bilateral testicular fixation.

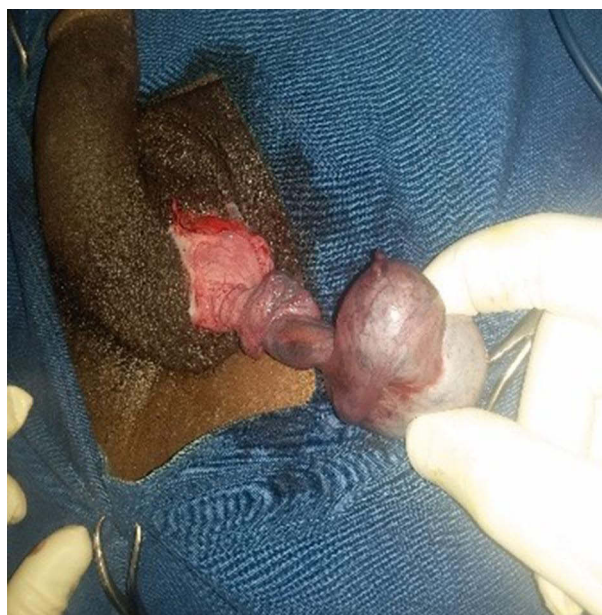


Figure 2 Operative findings of a torsion of the spermatic cord with three twists and a voluminous cephalic cyst of the epididymis.

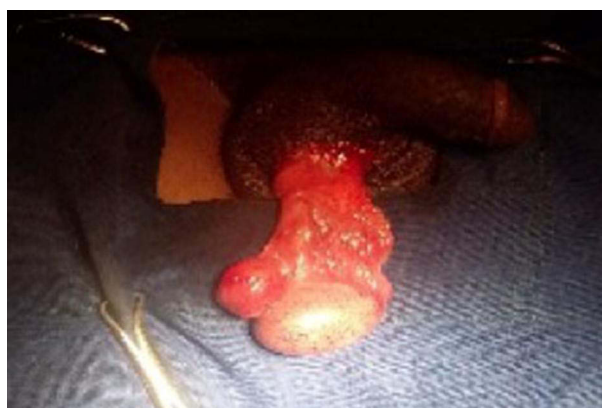


Figure 3 Progressive return to normal colour of the testis after detorsion.



Figure 4 Epididymal cyst after excision.

The post-operative course was uneventful, and the patient was discharged on the third day.

Clinical examinations and Doppler ultrasound at the sixth and twelfth postoperative weeks were both normal. A well-defined follow-up program was offered to him at the doctor's office, during which clinical and paraclinical tests would be performed (sperm analysis, hormone testing, and Doppler ultrasound), but the patient was lost to follow-up.

Discussion

Testicular torsion (TT) was first described by Hunter in 1810. It represents a substantial surgical emergency, as it jeopardizes the testicular functions and thus may affect fertility.^{10,12}

In a large majority of cases, it is caused by testicular tilting due to an anomaly in the testicular attachment system (long spermatic cord, absence of Gubernaculum testis, lax mesorchium) or a disproportion between the volume of the testis and its attachment systems.¹²

The latter condition can be indirectly fulfilled by an epididymal cyst. In fact, a large epididymal cyst, closely attached to the testicle, may become an integral part of it, increasing its weight. This creates a disproportion between the testicular weight and its attachments, with a greater risk for a rolling up around the spermatic cord.

Epididymal cysts are usually quite benign pathologies, requiring surgical treatment only in the case of painful cysts, such as for voluminous ones.¹³

In the case described in this manuscript, the voluminous epididymal cyst has tilted the testis by mechanical effect. This is supported by the lack of other classic TT factors in the patient: the spermatic cord was not excessively long, the tunica vaginalis of normal size, a well-outlined mesorchium and the scrotal ligament present.

In contrast, the classical obliquity of the testicular axis was obliterated by the cephalic epididymal cyst.

A "second testicle" sensation suggests an epididymal cyst, which is usually liquid-like and easy to distinguish from testicle tissue. Pain is often caused by cyst tension or torsion.¹⁴

The mild pain reported in our patient's history could be due either to the volume of the cyst or to the testicular subtorsion phenomena that often occur before TT itself.¹ The occurrence of severe scrotal pain in a patient who has had similar pain of lesser intensity should be interpreted as a diagnosis of TT. It indicates surgical exploration. This was the case in this study despite clinical and ultrasound findings that did not demonstrate torsional coils in the spermatic cord.

A large epididymal cyst may preclude the clinical examination of TT. The search for Gouverneur's sign (ascension and horizontalization of the testis) may be compromised by two factors: on the one hand, the testis has already lost the classic obliquity of its axis due to the superimposed epididymal cyst, and on the other hand, the cyst blocks the typical ascension of the gonad.

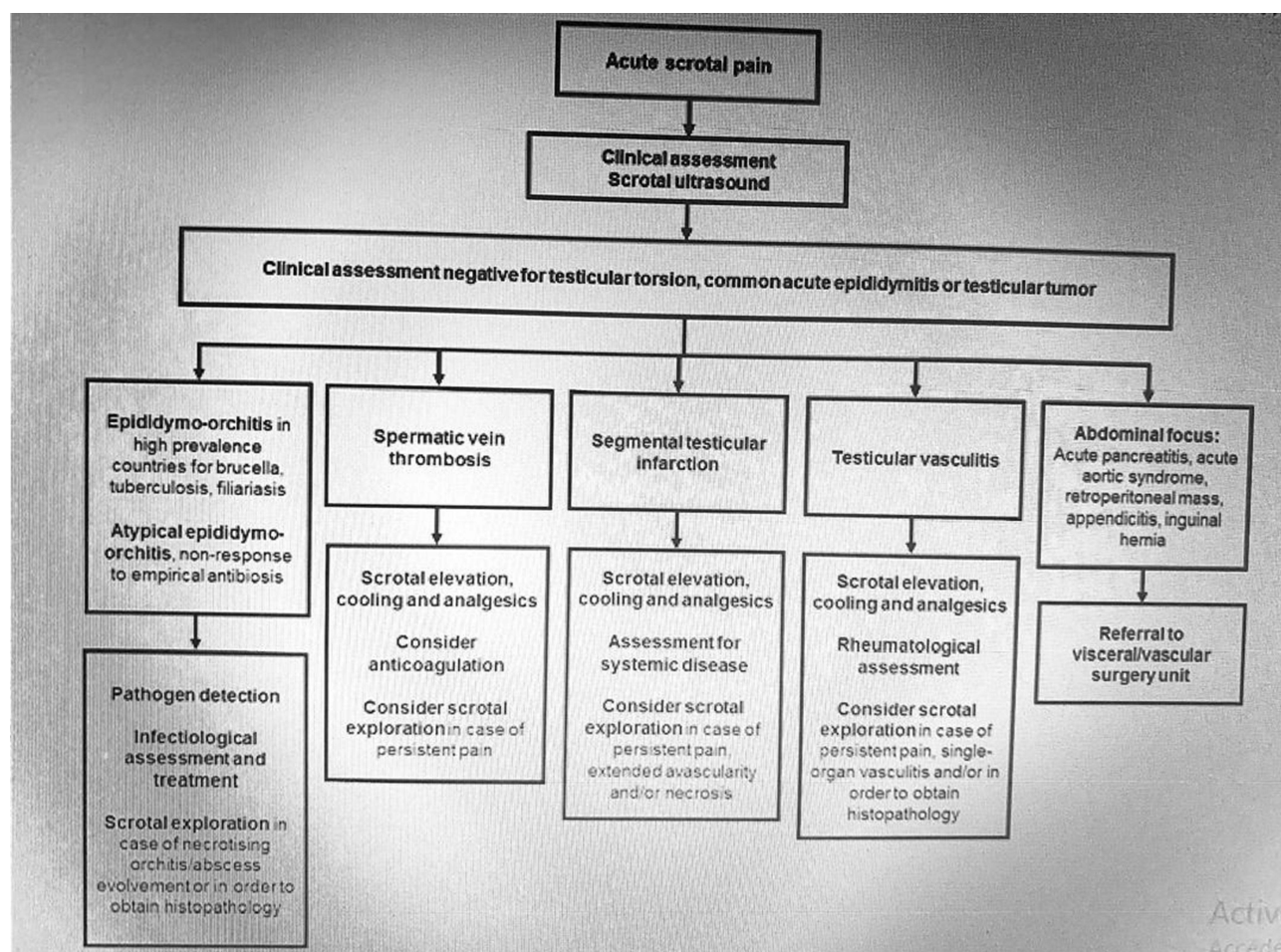


Figure 5 Differential diagnoses after exclusion of the most common causes of acute scrotal pain and treatment options as suggested by literature.

The search for torsion spirals is also greatly impaired by the presence of a large epididymal cyst due to the previa obstruction effect. There is also a real risk of mistaking a large cyst for a torsional coil in an edematous spermatic cord.

Although the pain is more severe in TT, it remains a subjective parameter.

Another difficulty is seen on ultrasound; a large epididymal cyst can hide the torsion coils. It is therefore easy to mix-up a big cyst and torsion coils in a swollen spermatic cord.

Nevertheless, it should be mentioned that no paraclinical imaging has been formally established to be useful in the definitive diagnosis of spermatic cord torsion.

Many cases of torsion of an epididymal cyst or testicular appendage without torsion of the spermatic cord have been described (Figure 5).¹⁵⁻¹⁹ On the other hand, TT on epididymal cysts are rare, with just one case reported in the literature to date, in a 57-year-old patient.²⁰

Weakness

Preoperative Ultrasound-Doppler images were not saved due to the emergency.

Conclusion

Although few or no studies in the literature have found a causal relationship between an epididymal cyst and TT, this observation leads us to consider epididymal cysts as a risk factor for TT. When monitoring patients with epididymal cysts, the risk of subsequent TT should be kept on mind.

In addition, a patient with a known epididymal cyst who reports severe scrotal pain should be operated promptly, as TT is not easily detectable under these conditions.

Compliance with Ethical Standards

This study received ethical clearance from the Ethical Committee of the Catholic University of Bukavu. We have obtained the approval of the Catholic University of Bukavu for the publication of this case report.

Consent

The patient provided written informed consent for publication of the case report and images.

Author Contributions

Data collection: BCW, LMB; Analysis and interpretation of results: CSM, DCM; Review of the article: LMM. All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the case report; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

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Disclosure

The authors declare no conflicts of interest in this work.

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