Torsion of spermatic cord in children: A review

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Abstract The current opinion on spermatic cord torsion is discussed in this review, with special attention to natural history, value of diagnostic tools, evidence for surgical management, outcome and management of atypical forms of torsion.
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Introduction
Acute spermatic cord torsion, more commonly and not so exactly named testicular torsion (TT), is one of the rare true emergencies in pediatric urology. Although there is no doubt about the necessity to restore impaired blood flow to the testis, there is still much debate with regard to the most effective diagnostic algorithm, indications for emergency surgery, management of the reperfused testis and management of less typical forms of TT (intermittent, late diagnosed and neonatal TT). We reviewed the current literature in an attempt to delineate an evidence-based strategy.

Incidence and pathophysiology
Testicular torsion occurs in approximately 1 in every 4000 men under the age of 25 years. The reported rate of TT in syndrome of the acute scrotum is variable. The most reliable data come from studies in which all children with symptoms of acute scrotum were indicated for surgical revision. In these, torsion of the appendix testis was the most common pathology (45–57%), followed by torsion of the spermatic cord (26–27%), and epididymitis as a much less common condition (only 10–11%) [1,2].

TT can be encountered at any age, but most commonly during adolescence and the neonatal period. Adolescent TT
is intravaginal. A long mesorchium allowing for greater mobility of the testis which hangs freely within the tunica space (bell clapper deformity) is believed to be the principal predisposing condition. This anatomical anomaly is nearly always bilateral. Hayn et al. found this abnormality in 47 boys who were operated upon for spermatic cord torsion or intermittent cord torsion in all cases on the affected side and contralaterally in 88% and 90%, respectively [3]. An autopsy study performed by Caesar et al. [4] revealed bell clapper deformity in 12% of the male population. As the incidence of spermatic cord torsion is much lower, an inciting event must occur. The sudden contraction of the cremasteric muscle, which is attached onto the cord in a spiral fashion, as a response to mechanical, sexual or thermic stimulation (e.g. a jump into cold water) may cause a rotational effect on the testis as it is pulled upward [5]. Perinatally, the mechanism of torsion is different. Absence of normal fixation between testicular coverings and tunica dartos results in abnormal mobility in the scrotum and extravaginal torsion.

The primary consequence of TT is ischemia of the testis due to venous and subsequent arterial occlusion followed by reperfusion injury upon repair. Hemorrhagic necrosis and cell apoptosis lead to the accumulation of testicular neutrophils (inflammatory response) and increased testicular oxidative stress. According to experimental animal studies, germ cells are much more vulnerable than Sertoli cells and spermatogenesis can be eliminated despite returned blood flow and preserved Sertoli cell function [6]. However, the exact mechanism of cellular injury remains only partially understood.

The two most important factors determining testicular damage are the length of time from onset of symptoms to cord detorsion, and the degree of cord twist. It is commonly accepted that salvage of the human testis declines sharply with a symptom duration greater than 6 h [7]. However, necrosis is possible as early as within 4 h where a high degree of twisting exists [8]. In animal studies, blood flow changes in testicular tissue were insignificant during torsion and after detorsion if the cord was twisted to 360° or less. In the 720° torsion group, blood flow to the twisted testis was reduced significantly [9]. Tryfonas et al. found complete or severe atrophy in all patients with torsion of more than 360° and a symptom duration of more than 24 h [10].

Salvage rate of testes after detorsion is reported as 62–85% for descended testes and 29–40% for undescended testes [11]. The majority of authors report the testis as ‘salvaged’ if a palpable mass with Doppler signs of perfusion is preserved. However, this does not correspond to salvaged testicular function. Although, duration of ischemia correlates well with testicular atrophy, there is no clear correlation with semen quality. Semen analysis is normal in only 5–50% of patients [12], indicating alteration of the contralateral testicle. There are many theories explaining this ‘collateral damage’. The theory of an autoimmune process triggered by the rupture of the hematotesticular barrier leading to formation of anti-sperm antibodies has never been confirmed. Evidence against an autoimmune reaction is found in the article by Anderson et al. [13], who did not detect any cases of anti-sperm antibodies after TT. Hagen et al. [14] found pre-existing dysplastic changes in the contralateral testicle in 30 of 34 patients (desquamation of the germinative epithelium, atrophy of the Leydig cells and malformation of spermatoblasts). This may represent consequences of reperfusion injury episodes during previous asymptomatic intermittent torsion or congenital abnormality. Pre-existing contralateral abnormalities were verified by Anderson et al. [13]. Altered microcirculation of the opposite testis due to vasospasm caused by a sympathetic mediated reflex may affect testicular function and ultimately fertility as well [15].

### Incidence and pathophysiology

- Torsion of the spermatic cord represents 26–27% of children with acute scrotum.
- Two most important factors determining testicular damage are time from onset of symptoms to cord detorsion and degree of cord twist.
- Poor semen quality after testicular torsion may represent consequences of previous symptomatic intermittent torsion or congenital abnormality.

### Diagnosis

#### History and clinical findings

The goal of diagnosis is to identify boys with TT as soon as possible. Rapid diagnosis shortens the duration of ischemia; correct diagnosis lowers the incidence of unnecessary surgery. Acute and rapidly increasing pain is the most common symptom. It may be localized not only in the scrotum but also into the groin and lower abdomen, imitating an acute abdominal event. It may occur during sleep and arousal of the patient; it may follow direct trauma (in 4–8% of cases), increased physical activity or sudden change of temperature (dive into cold water). Some patients refer to intermittent acute pain, uni- or bilateral, with spontaneous resolution in their history. Vegetative symptoms like nausea, vomiting, paralytic ileus, lethargy may be encountered. In neonates, signs of restlessness may be associated with the torsion, but usually the first sign is a scrotum of abnormal appearance during delivery or diaper changing.

On early examination, painful palpation and scrotal edema are found in 88% and 44% of patients, respectively [2]. Skin redness and elevated body temperature are usually absent. Painful enlarged testicle and epididymis are typical early signs; the testicle is placed more cranially due to the shortening of the cord. After several hours the scrotal skin becomes red; the massive local swelling makes clinical examination difficult, sometimes nearly impossible. Clinical examination of the abdomen is normal. Cremasteric reflex is a very useful diagnostic tool. Rabinowitz [16] described 100% correlation between the presence of the ipsilateral cremasteric reflex and the absence of testicular torsion during a 7-year evaluation of 245 boys with acute scrotal swelling. Later, one case of preserved cremasteric
reflex accompanying spermatic cord torsion was described in the literature [17]. The specificity of the cremasteric reflex is low. Attenuation or absence of reflex is found in up to 34% of healthy boys. In a recent and unique prospective study identifying parameters of history and clinical findings and using multivariate logistic regression analysis, absence of ipsilateral cremaster reflex, nausea/vomiting and scrotal skin changes were the only consistent predictive factors of testicular torsion [18].

As there is an overlap of signs and symptoms of spermatic cord torsion with other conditions (acute epididymitis, epididymo-orchitis, torsion of appendix testis, strangulated hernia, testicular trauma etc.), the clinician may not make an accurate diagnosis according to the history and clinical examination only. There are several methods available to help the clinician, who is limited by the urgency of the condition. Urine dipstick examination is useful to diagnose canalicular epididymitis and is normal in TT.

**Ultrasonography**

Ultrasonography represents the basic and most useful diagnostic tool. Its strong points are: good accessibility, rapidity, non-invasiveness, and capacity to show anatomic details valuable for differential diagnosis. It is crucial to realize that just as the intensity of the clinical findings changes with time, the ultrasonographic findings vary according to the duration of ischemia. Currently, several ultrasonographic methods are being used.

Gray scale imaging demonstrates an enlarged testis, most frequently with diffuse hypoechogenicity and sometimes good identification of lobar architecture (interstitial and septal edema) of the affected testis. Diffuse multifocal hyper- and hypoechogenicities correspond with hemorrhagic infarction. The epididymis is enlarged with a heterogeneous echo texture. Thickening of peritesticular tissues due to edema is gradually increasing. However, for the first 2–4 h, sonographic appearance may be normal [19]. Cranially to the testis, a large echogenic extratesticular mass that contains the edematous part of the epididymis and twisted segment of the cord may be identified. With the help of a high-frequency transducer (HRUS: high resolution ultrasonography), identification of the exact site of torsion and the spiral aspect of the twisted spermatic cord (whirlpool or snail shell-shaped) is possible. A multicentric study from 11 European university hospitals resulted in efforts to develop a new modality for highly sensitive quantitative evaluation that is less dependent on the expertise of the investigator. New methods include contrast-enhanced ultrasound (CEUS), perfusion CT (PCT), dynamic contrast-enhanced magnetic resonance imaging (CE-MRI), and near-infrared spectroscopy (NIRS). CEUS, PCT and CE-MRI are perfusion studies performed by analyzing the first pass of contrast agent bolus through the vasculature that causes a transient increase in detected signal in the healthy testis compared with the affected one.

CEUS is easy to perform in emergency clinical settings. After baseline ultrasonography, a bolus injection of ultrasound contrast agent (USA) is administered intravenously to the systemic circulation. The USA is formed of gas-filled microbubbles having a high degree of echogenicity, thus improving the depiction of parenchymal vascularity. The power of the ultrasound beam (mechanical index) is set at the minimum and the focus placed at the end of the field of view to obtain the minimum of microbubble destruction. A recent study by Valentino et al. demonstrated that CEUS can lead to correct diagnosis of TT in cases where color Doppler ultrasound is inconclusive [25]. A promising future method upgrade is contrast-enhanced, real-time volumetric ultrasound imaging, which enables quantitative analysis of global perfusion of the testes; only an animal study is available so far [26].

Experimental studies have demonstrated good sensitivity of PCT in the early detection of testicular ischemia...
However, the main disadvantage of this method is the exposure of gonads to higher radiation.

MRI is a highly specific and sensitive imaging modality for evaluation of scrotal pain. The conventional T1 and T2 weighted images can detect TT by distinguishing the whirlpool pattern and the knot in the spermatic cord. When MRI is performed with contrast enhancement, ischemia caused by torsion of a testicle can be detected. Watanabe et al. [28] examined 14 patients with TT using dynamic CE-MRI and reported an outstanding 100% sensitivity in cases of complete torsion in contrast to 75% sensitivity for T2-weighted imaging. A recent animal study of diffusion-weighted MRI has shown its feasibility to detect TT without the use of contrast media [29]. Unfortunately, attaining an urgent MRI study by an experienced radiologist may be difficult; younger children need general anesthesia and the investigation is more time consuming than ultrasonography. This decreases its present value in clinical practice, but as an alternative method it should be taken into consideration.

NIRS does not detect tissue perfusion, but reveals oxygen saturation, which is more vital than blood flow. In a recent animal study, NIRS correctly identified all testes where color Doppler ultrasound was inconclusive. CEUS may lead to correct diagnosis of TT in cases where color Doppler ultrasound was inconclusive. [30]. It is a promising candidate for fast and cost-effective initial evaluation of TT in the emergency setting. At present, there is only one recent human study available [31].

**Diagnosis**

- Typical history and typical clinical findings necessitate prompt scrotal exploration and no auxiliary imaging is indicated.
- Symptoms, signs and ultrasound view develop and change over time according to the duration and severity of ischemia.
- Early on, the only symptoms may be painful enlarged testicle and epididymis; redness and edema of skin appear later; sonographic appearance may be normal within the first hours.
- Positive finding of ipsilateral cremasteric reflex strongly correlates with absence of TT.
- Doppler ultrasound has variable sensitivity dependent on investigator’s experience; HRUS of spermatic cord is helpful.
- Absence of detectable flow in the affected epididymis may help confirm the diagnosis.
- CEUS may lead to correct diagnosis of TT in cases where color Doppler ultrasound is inconclusive.
- NIRS is a promising candidate for the initial evaluation of TT.

**Treatment**

According to the EAU and ESPU guidelines [32], an urgent exploration is indicated within the first 24 h from the onset of symptoms. After 24 h, semi-elective surgery may be adopted (level of evidence 3; grade of recommendation C).

However, from the history, we cannot assess the degree of cord twist and also the duration of significant ischemia may be doubtful. Therefore, if the clinical and ultrasound findings are not reliable enough to estimate the residual vitality of the testis, an urgent revision, even if the patient presents late, should be adopted.

**Manual detorsion**

Preoperative manual detorsion is the fastest way to release ischemia. Unfortunately, the traditional thinking that TT occurs primarily in the medial direction is misleading, since it occurs in the lateral direction in a third of cases [33]. Pulling the testis down, detorsion should first be executed in the lateral direction. If the patient does not experience an immediate release of pain, it should be executed in the opposite direction. Cornel et al. [34] were successful in 14 of 17 patients with cord torsion. However, manual detorsion of the spermatic cord is not a substitute for surgical exploration and bilateral orchidopexy. Sessions et al. found residual torsion in 32% of patients after manual detorsion [33].

**Surgical revision**

The testicle is exposed through a small horizontal incision on the anterolateral aspect of the scrotum. In the majority of cases, detorsion is directed laterally. If the blood supply is restored and the testicle gains adequate color, it is fixed with two to three non-absorbable sutures to the raphe scroti. Kuntze et al. found in a review that an absorbable suture was primarily used in 15 of 16 patients with recurrent torsion [35]. The patients should be informed that recurrent torsion following testicular fixation may occur many years after the primary procedure. Mor et al. [36] described 8 recurrent torsions in his group of 179 patients with fixed testes. Average time to recurrence was 7 years. Animal studies showed that testes fixed with non-absorbable material were adherent only at the sutures, while darts-fixed testes demonstrated complete circumferential adherence [37]. Therefore, some authors recommend partial resection or eversion of the tunica vaginalis resulting in extensive superficial adherences as a more effective and reliable method to prevent recurrent torsion.

Because the anatomical anomaly (bell clapper deformity) is usually bilateral, fixation of the contralateral testis should be performed, ideally at the same session. If the testicle does not gain appropriate color after detorsion and necrosis is obvious, orchidectomy is performed together with the contralateral orchidopexy. Autoimmune injury to the contralateral testis has not been proved and the andrological prognosis is similar in patients with a preserved testicle following torsion and those with the testicle removed [38].

**Treatment of ischemic-reperfusion injury**

With increasing knowledge concerning pathogenetic pathways responsible for ischemic-reperfusion injury, protective measures to decrease the impact of oxidative stress have been studied; although many molecules with
a potential positive effect in animal studies have been described, none of them have been confirmed in humans. Another theoretical way to minimize ischemic-reperfusion injury is to prevent intratesticular compartment syndrome (analogous to compartment syndrome in vascular surgery). The goal is to lower the pressure in edematous testicular tissue enclosed in firm tunica albuginea. In rats, Moritoki et al. [39] showed a significant correlation between intratesticular pressure and spermiogenesis 4 weeks after detorsion. Testicular capsulotomy after detorsion can release testicular compartment syndrome effectively in humans [40]. However, animal studies did not confirm the positive effect of capsulotomy on testicular histology [41] and functional results are not available in humans.

Less common manifestations of torsion of the spermatic cord

Intermittent torsion

History of recurrent self-limiting attacks of strong scrotal pain irradiating into the groin should alert the clinician to the diagnosis. Nausea and/or vomiting have been reported in a quarter of patients [42]. At time of examination, the clinical findings are usually normal. Horizontal position of the testes in the upright position is a highly sensitive marker of the bell clapper deformity. Eaton et al. [42] described 100% correlation at surgical exploration. Due to the transient character of testicular ischemia, color Doppler ultrasonography is of little help. If intermittent spermatic cord torsion is a likely diagnosis, patients should be informed about the condition and that bilateral orchidopexy needs to be performed.

Late presented torsion

Adolescent timidity, partial alleviation of symptoms and initial misdiagnosis may delay the correct diagnosis. In these subacute cases, the pain is usually absent or minimal. Local scrotal swelling and redness are much less marked. At palpation, the testicle is large, firm and minimally painful. The testis and epididymis can hardly be differentiated. As the exact duration of ischemia is difficult to determine (possible intermittent ischemia), the principal question is whether to perform an urgent scrotal exploration in a patient presenting with a history slightly longer than 24 h. The decision should be based on clinical findings (dead tissue is not painful) and the ultrasonographic appearance. Kaye et al. [43] found that heterogeneity on preoperative ultrasound was universally predictive of organ loss in all 37 patients of his group, whereas 89% of patients demonstrating a homogeneous and isoechogenic image were deemed viable at exploration. Thus, in the setting of Doppler proven TT, a heterogeneous parenchymal echo texture indicates testicular non-viability. This conclusion was recently confirmed by Chmelnik et al. [44] for all age groups. Therefore, if a heterogeneous parenchymal echo texture is found, elective scrotal exploration may be adopted; emergency exploration is indicated if a homogeneous echo texture is present.

Torsion of the intraabdominal testis may manifest under a clinical picture of acute abdomen. Torsion of the inguinal undescended testis may imitate an incarcerated hernia. Therefore, in the case of a patient presenting with abdominal pain and having an impalpable testis, TT must always be considered. The rates of surgical salvage are low, which is mainly attributed to delay in parental response and to diagnostic error resulting in delay in establishing the correct diagnosis.

Perinatal testicular torsion

Perinatal testicular torsion (PTT) is defined as torsion of the spermatic cord that has happened prenatally, perinatally or postnatally up to the 30th day of life. Since first reported in 1897 [45], torsion of the neonatal testis has become well recognized, and with its incidence of 6 per 100,000 live births [46] represents about 10% of all cases of torsion. Several controversies regarding management of this condition still persist. Current management strategies include observation alone, and postponed or emergency contralateral orchidopexy. Proponents of the wait and see policy refuse scrotal exploration and advise parents to examine the scrotum at each diaper change. They argue that prenatal (in utero) TT is never salvageable, asynchronous bilateral torsion is extremely rare, and general anesthesia in the newborn carries a major risk [47,48]. The opponents state that physical examination and radiography can be inaccurate to assess the state of the contralateral testis. Since the possible loss of the remaining testicle would be a catastrophic event, emergency surgical exploration should be adopted to protect the healthy gonad [49].

We believe the confusion has arisen because many authors do not distinguish between the different entities of PTT. In 1990, Das and Singer [50] proposed PTT to be classified as ‘in utero torsion’ (prenatal and perinatal) and ‘postnatal or neonatal torsion’ according to the...
clinical findings immediately after birth. In utero torsion is represented by a firm, non-transilluminable scrotal mass, mostly with bluish or no discoloration of scrotal skin, that is painless (long-lasting ischemia has already damaged sensoric tissue innervation). The postnatal torsion is characterized by a red, swollen, painful scrotum that was clinically normal at the moment of birth. Sorensen et al. pointed out the practical impact of this nomenclature on the fate of different testicles [51], presenting four salvaged testicles in a group of 10 cases with evident postnatal torsion. For clarification of this issue, we reviewed the literature cited in Medline from 1990 to 2011, looking for the fate of the testis in the case of in utero and postnatal torsion. However, many reports did not make a distinction between these two principal entities. In many cases, clear information about clinical findings at delivery was overlooked and not evaluated. Additionally, many reports are only isolated cases and the authors vary widely in management strategies. Among 321 cases of PTT, we were able to identify 25 cases of postnatal torsion and 141 cases of in utero torsion. In the remaining 155 cases, we could not find sufficient information to decide on the character of perinatal torsion. From the 25 postnatal torsions, 12 testicles (48%) were salvaged. This would support urgent management of all cases with postnatal/perinatal torsion. On the other hand, all testicles affected prenatally became atrophic with an exception of two cases (0.7%). Al-Salem described an unusual case of a salvaged testis after a 30-h history of torsion [52]. Sorensen et al. found a case of a newborn, recorded immediately at birth, with an enlarged soft left hemiscrotum and normal looking right side. Scrotal exploration carried out 3 h later revealed bilateral torsion. The left-sided testicle had been affected recently and was salvaged, the right-sided testicle, probably affected much earlier, was grossly necrotic and was lost. We might conclude that prenatal torsion without obvious acute changes is probably an unsalvageable event [53]. Nevertheless, we have found more reasons in favor of early scrotal exploration of the prenatal torsion.

Firstly, asynchronous torsion does exist and carries an immediate risk of bilateral anorchia. The only measure to prevent this disaster is early contralateral testis fixation. The wait and see course may be dangerous since the risk of possible asynchronous torsion should lead us to schedule the newborn to ipsilateral orchidectomy and contralateral orchidopexy as soon as possible. In the case of in utero torsion, presenting with a heterogeneous parenchymal testicular echo texture, emergency bilateral exploration is able to distinguish between these two types of torsion.

Secondly, contralateral asymptomatic PTT has been repeatedly reported during scrotal exploration, when preventive fixation was being attempted. Baglaj et al. reported only 6 of 16 patients with bilateral torsion in whom diagnosis of torsion of the contralateral testis was made preoperatively [54].

We can theorize that in the case of late presentation the local inflammatory reaction has already been healed. The sensitivity of color Doppler ultrasound in identifying an absent flow is of limited value in newborns. The examination is challenging even for an experienced radiologist, and the findings can be inaccurate and do not exclude ischemia, especially when the interruption of testicular blood flow is partial. In the literature, several authors have reported a necrotic testis in spite of normal Doppler scans preoperatively [46,54].

Therefore, the principal role of ultrasound in newborns is to visualize the architecture of the scrotal content to evaluate for the differential diagnosis (tumor, hematoma, inguinal hernia, and meconium periorchitis) and to detect the presence of testicular necrosis. A heterogeneous parenchymal echo texture is typical for tissue necrosis and can predict testicular loss with high sensitivity [43]. In cases of in utero torsion, presenting with a heterogeneous parenchymal testicular echo texture, when salvage of the affected testicle cannot be expected, the risk of possible asynchronous torsion should lead us to schedule the newborn to ipsilateral orchidectomy and contralateral orchidopexy as soon as possible. In the case of in utero torsion, presenting with a homogenous parenchymal echo texture, emergency bilateral exploration should be done.

Fourthly, current opinion concerning the significantly elevated risk associated with general anesthesia in newborns should be challenged as well. The majority of infant mortality statistics are drawn from seriously ill patients with life-threatening conditions such as congenital cardiac and gastrointestinal malformations. In contrast, nearly all boys affected with PTT are full term, otherwise healthy infants [58]. The frequently cited study of Warner et al. reporting increased anesthetic complications in young infants with herniorrhaphy is outdated since the quality of postoperative care has much improved in the last two decades [59]. In our literature review covering explorations of PTT during the last 21 years, we have not encountered any report of a life-threatening complication. The evidence presented arguments it is hard to justify a passive approach to the condition, potentially resulting in bilateral testicular atrophy. This is even more applicable in a contemporary litigious society.
Less common manifestations of torsion of the spermatic cord

- History of recurrent self-limiting attacks of strong scrotal pain associated with horizontal position of the testes in the upright position makes diagnosis of intermittent spermatic cord torsion highly probable
- In the setting of doppler proven late testicular torsion, heterogeneous parenchymal echo texture indicates non-viability of testicular tissue in all age groups
- In the case of a patient presenting with abdominal pain and having an impalpable testis, torsion of cryptorchid testicles should always be considered
- In newborns, clinical and Doppler signs are not reliable in excluding asynchronous torsion; bilateral surgical exploration is recommended in all cases of perinatal torsion.

Conclusions

There is no doubt that testicular ischemia resulting from spermatic cord torsion should be released as soon as possible to prevent irreversible tissue damage. The key to correct differential diagnosis and decision is the knowledge that symptoms, signs and ultrasound view are developing and changing over time according to the duration and severity of ischemia.

Doppler ultrasound has variable sensitivity significantly dependent on the investigator’s experience and can be enhanced with the help of high resolution ultrasonography.

Detorsion remains the only therapeutic procedure. There is not enough evidence to use adjuvant measures to influence ischemic-reperfusion injury after detorsion in humans.

In newborns, clinical and Doppler signs are not reliable to exclude asynchronous torsion. Therefore, bilateral surgical exploration is recommended in all cases of perinatal torsion.

Conflict of interest

The authors have not declared any conflict of interest.

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