

# Is testicular torsion a real problem in pediatric patients with cryptorchidism?

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## Research

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# Abstract

## Purpose

To present management and outcomes of patients with cryptorchidism suffering from testicular torsion.

## Methods

This is a retrospective review of pediatric patients with torsion of undescended testes, who were treated between 2009 and 2019. On the medical charts, we recorded: the age, symptoms, duration of torsion, physical examination, surgery findings, and additional treatment.

## Results

We identified 9 boys with torsion of the undescended gonad, which represented 4.7% of all 192 boys with testicular torsion. The mean age of boys with torsion of undescended testis was 8.7 years. The mean duration of symptoms in our study group was long and it was up to 28.5 hours. All patients had inguinal canal exploration. In 8 cases, testicular necrosis and primary orchidectomy was performed.

## Conclusion

According to clinical experience and available studies, torsion of male undescended gonads is a comparatively rare condition. Nevertheless, diagnosis is still delayed and connected with inevitable orchidectomy.

## Plain English summary

Authors present 9 cases of boys with torsion of undescended testes. They represented 4.7% of all boys with testicular torsion treated during 10 years in Pediatric Surgery Department. The mean age of boys with torsion of undescended testis was 8.7 years. The mean duration of symptoms was long and it was up to 28.5 hours. All patients were operated on, in 8 cases necrotic testicle was removed. Even though torsion of undescended testicle is quite rare, diagnosis is delayed and because of that, boys are subjected to removal of the testicle.

## 1. Introduction

Acute scrotum is a medical emergency. In the pediatric population, the most common causes of this pathology are usually: hydatid or testicular torsion and epididymo-orchitis (1, 2). The main symptoms are most commonly: scrotal pain, swelling and redness of the scrotum. Nevertheless, testicular torsion is the most serious cause of acute scrotum and may result in the loss of the testicle. Testicular torsion could be extra- or intravaginal. Usually extravaginal is typical for the neonatal period and includes the testicle, epididymis and tunica vaginalis. Contrary to extravaginal, intravaginal torsion is connected with a bell-

clapper deformity. In this abnormality, the mesorchium terminates early and the testis is free floating in the tunica vaginalis.

Cryptorchidism is the most common genital disorder in males (3), and generally should be diagnosed and treated in the neonatal period (4). The consequences of an undescended testicle are higher risk of testicular cancer (5) and impaired fertility (6). Moreover, cryptorchidism is connected with a higher risk of gonadal torsion (7–9). Because of the risk of testicular necrosis, torsion of the gonad is the most urgent disorder and surgical emergency. Therefore, prompt diagnosis and urgent surgical treatment are crucial.

Testicular torsion can usually be diagnosed based on a careful physical examination and appropriate Color-Doppler Ultrasound (CDU). However, one should consider the fact that physiologically in prepubertal boys and neonates intratesticular blood flow is reduced, which may result in false-positive ultrasound results. Correct interpretation seems to be more difficult in the case of incorrect placement of the gonads (undescended testis –inguinal and especially abdominal undescended testis). Approximately 80% of undescended testicles are palpable (3) and located in the inguinal canal. Physical examination is a basic technique in the diagnosis of cryptorchidism.

In this study, we analyzed patients with torsion of undescended testis.

## **2. Materials And Methods**

In this retrospective study, we analyzed all boys with testicular torsion, who were operated on in the Pediatric Surgery and Urology Department of the Medical University of Bialystok (Poland) between 2009 and 2019. On the medical charts, we: the age, symptoms, duration of torsion, physical examination, surgery findings and additional treatment. Patients with torsion of undescended gonads were selected and included in the study.

## **3. Results**

Over the last 10 years (from 2009 to 2019), 192 boys with testicular torsion were operated on. In this group, we identified 9 boys with torsion of the undescended gonad, which represented 4.7% of all boys with testicular torsion. Six patients had torsion of the left testicle (66.7%).

The described 9 patients with torsion of undescended gonads accounted for 0.95% among 860 boys operated on in a 10-year period because of cryptorchidism (congenital and ascending gonads).

The mean age of boys with torsion of undescended testis was 8.7 years (range 6 months – 14 years).

Among our patients, 6 had general good health, 3 other subjects suffered from cerebral palsy, and 1 had epilepsy.

The most common complaints in all patients were local symptoms: painful inguinal swelling and redness of the groin. Additionally, we noticed general manifestations like abdominal pain, vomiting and fever.

The mean duration of symptoms in our study group was 28.5 hours (range 6 h-4 days). Only one patient was admitted to the hospital during the first 6 hours from the onset of symptoms. In all cases, a diagnosis of cryptorchidism was made. In 3 patients bilateral cryptorchidism was diagnosed at the age of 10, 11, and 13 years, respectively. Interestingly, these 3 boys with cerebral palsy had their diagnosis of cryptorchidism made a few years earlier. Nevertheless, caregivers did not decide to have their children operated.

Urgent Color-Doppler Ultrasound was performed before operation in all boys. In 2 cases (22.2%), proper diagnosis had been made. In one patient, preoperative CDU suggested enlarged lymph node. In the CDU of 3 boys, inflammation of inguinal testis and epididymis was suspected. In 2 cases, a diagnosis of appendage torsion was made, and in 1 case, after CDU examination, suspicion of incarcerated inguinal hernia was made.

Regardless of the results of the CDU examination, all patients with the symptoms of torsion underwent urgent surgical inguinal region exploration. In one case of a 6-month neonate, extravaginal torsion was detected. In other cases, the intravaginal form of torsion was found. In 8 cases of boys with testicular torsion, we found testicular necrosis, and primary orchidectomy was performed. Figure 1. In 1 case, during surgery we noticed proper reperfusion of the gonad and orchiopexy was performed. Unfortunately, this patient did not return for follow-up. In all cases, fixation of the other testis was done. In the postoperative period, all boys were given intravenous antibiotics (cefuroxime-7 patients, amoxicillin – clavulonate-2 patients). We did not notice any postoperative complications, like wound infections or fever. Table 1.

## 4. Discussion

The first case of testicular torsion was described in 1840, by Delasiauve (10). It was a teenager with cryptorchidism, and orchidectomy of necrotic testis was performed. Nevertheless, the incidence of torsion in cryptorchid patients is still unknown. Generally in the literature, descriptions of a series of cases can be found (9, 11–13). There are no randomized trials with unambiguous guidelines. In 16 years, Naouar et al. (14) found 13 cases of torsion of undescended testicle, 1 adult and 12 children. Similarly, other authors (15) presented a two-center review of 11 patients with torsion of undescended testicle, who account for 9.7% of all boys with testicular torsion. In our study, 4.7% of patients with testicular torsion suffered from cryptorchidism.

The mechanism of testicular torsion of the undescended testis is not well understood (12). There are some theories about the pathophysiology of torsion of the undescended testis. Incorrect spasm or contraction of cremasteric muscle is one of them (12, 14). Additionally, in children with cerebral palsy contracture of the hips could be a potential risk factor (16). A recent study suggested that the prevalence of cryptorchidism in patients with cerebral palsy is approximately 10-fold greater than in the general population (17). It is also strongly associated with spastic quadriplegia. In our series, we had 3 patients with cerebral palsy. Another risk factor is an increased size of the gonad in patients with testicular tumors

(8). We did not find any cases of tumor in our patients. On the other hand, similarly to scrotal torsion, we found that the extravaginal type was recognized only in a 6-month-old neonate. There are 2 peaks in the incidence of testicular torsion in the pediatric population: during the first year of life and from 12 to 18 years (18, 19). In our patients, we also observed the same age - dependency.

Clinical signs are less obvious than usually. We know that acute scrotal pain, especially connected with additional clinical signs is an emergency and requires rapid medical consultation. Pain or swelling in the inguinal canal are not so alarming. In this kind of patient we should differentiate between incarcerated hernia, appendicitis, lymphadenitis, renal colic or groin injury (20). As shown in our study, patients suffer from nonspecific complaints like fever, vomiting or abdominal pain. It should be emphasized that a meticulous physical examination (including inguinal canal and scrotum area) is crucial and increases the chance of saving the testicle.

Imaging tests are useful for a proper diagnosis. Ultrasound examination is a safe, accessible and fast tool to make a proper differential diagnosis. In different reports, the sensitivity of color Doppler ultrasound ranged from 69.2 to 100% (2, 21, 22), although it could be challenging and depends on the experience of the physician. In our study, only in 22.2% of the cases the a correct ultrasound diagnosis has been made. Regardless of the results of the CDU, all patients with symptoms of acute scrotum and suspicion of testicular torsion were qualified for surgical intervention.

Testicular salvage is possible when a surgical intervention is performed within the first 6–8 hours from the start of symptoms (23, 24). According to a systematic review (25), when the spermatic cord is untwisted within 6 hours 97.2% of the testis survives. In our series, the time to surgical intervention was much longer. It seems that one of the reasons is low awareness of the caregivers. Friedman et al. showed that only 34% of parents are aware of this condition (26). According to the literature, one-third of cases of scrotal torsion ended with orchidectomy (27, 28).

What seems to be important, our case series also shows that treatment of patients with cerebral palsy with coexisting cryptorchidism is an unsolved ethical dilemma (29). According to the Nordic recommendation, orchiopexy should be performed between the 6th and the 18th month of life (30). Nevertheless, there is an ethical discussion about patients with neurological disorders. Usually, caregivers' wishes regarding treatment options are taken into account. It appears that especially nonpalpable and high positioned testes are associated with a potentially higher risk of the negative consequences of this disorder (31). Therefore, the positioning of the gonad in the subcutaneous area in the inguinal canal enables further control. Furthermore, it is highly recommended to combine orchiopexy with other surgical procedures (orthopedic, etc.). We should also bear in mind the possibility of intra-abdominal testicular torsion (32–34). Usually, the most common symptom is abdominal pain. In the literature we can find cases of testicular torsion of the intra-abdominal seminoma (35, 36).

## 5. Conclusion

According to clinical experience and the available studies, torsion of the male undescended gonad is a comparatively rare condition. Nevertheless, male patients with inguinal swelling and tenderness should be carefully and urgently examined. All patients need surgical/urological consultation. As it is presented in our research, diagnosis is still delayed and connected with the inevitable orchidectomy. Both physicians and caregivers should be aware of possible torsion of the inguinal testis to increase the possibility of saving the gonad.

## Declarations

Ethics approval and consent to participate:

This study was accepted by the Ethics Committee of Medical University of Bialystok Poland

Consent for publication:

all the parents/caregivers of our patients gave their informed consent for the study.

Availability of data and materials:

The datasets analysed during the current study available from the corresponding author on reasonable request

Competing interests:

The authors declare that they have no competing interests

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Authors' contributions:

MK, AP, EM - conception and design, acquisition of data, analysis and interpretation of data, drafting of the manuscript, AH - acquisition of data, analysis and interpretation of data, WD - revising of the manuscript All authors read and approved the final manuscript

All authors certify that the manuscript is a unique submission and is not being considered for publication by any other source in any medium. The manuscript has not been published, in part or in full, in any form.

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## Figures



**Figure 1**

Extravaginal inguinal testicular torsion in a 6 month-old boy.