Cryptorchid testicular torsion in children: characteristics and treatment outcomes

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This study aimed to review and compare the characteristics and treatment outcomes of cryptorchid testicular torsion in pre- and postpubertal children. We reviewed the clinical data of 22 patients with testicular torsion complicated by cryptorchidism who were treated between January 2010 and December 2019. Patients were categorized into prepubertal (1 month to 9 years; n = 12) and postpubertal groups (10–16 years; n = 10). The age at presentation, clinical presentations, physical examination, and operation outcomes were assessed. The common clinical presentations in both groups were inguinal pain and a tender inguinal mass. Patients in the prepubertal group were significantly more likely to present with restlessness (33.3%) than those in the postpubertal group (0%; P = 0.044). After detorsion, testicular blood flow recovered during surgery in 25.0% of the prepubertal and 80.0% of the postpubertal patients (P = 0.010). Orchiectomy was required in 50.0% of the prepubertal and 20.0% of the postpubertal patients (P = 0.145). Of the 22 patients with follow-up data, the rates of testicular salvage were significantly different, at 16.7% in the prepubertal patients and 60.0% in the postpubertal patients (P = 0.035). Cryptorchid testicular torsion has various manifestations. Although an empty hemiscrotum and a painful groin mass were common in both groups, restlessness was more prevalent in the prepubertal patients and 60.0% in the postpubertal patients (P = 0.035). Cryptorchid testicular torsion has various manifestations. Although an empty hemiscrotum and a painful groin mass were common in both groups, restlessness was more prevalent in the prepubertal patients during early testicular torsion onset than that in the postpubertal patients. Notably, the testicular salvage rate was significantly lower in the prepubertal patients than that in the postpubertal patients.

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INTRODUCTION

Testicular torsion is a common pediatric urologic emergency that requires immediate attention and treatment. The annual incidence of testicular torsion is 3.8 per 100 000 males younger than 18 years.1 If treated within 6 h of the presenting pain, 90%–100% of the testes can be saved. If treated within 6–12 h, depending on the degree of torsion, 20%–50% of the testes can be rescued, and if treated within 12–24 h, only up to 10% of the testes can be saved.2 After 24 h, the affected testis is likely to require orchiectomy, which can subsequently result in decreased fertility and hormonal function.3

The typical symptom of testicular torsion is scrotal pain, but in cases for which the twisted testicle is not located in the scrotum, such as in those with undescended testes (UDT) or cryptorchidism, the diagnosis becomes more difficult for a physician. The occurrence of cryptorchid testicular torsion in children is very rare, and parents or caregivers may not consider this condition a true surgical emergency. Therefore, presentation to a medical professional and a correct diagnosis may be delayed, especially for prepubertal children who cannot describe their symptoms on their own. This study aimed to compare the characteristics and outcomes of testicular torsion complicated by cryptorchidism between pre- and postpubertal patients. We hypothesized that the manifestations and outcomes of cryptorchid testicular torsion in children are different before and after puberty.

PATIENTS AND METHODS

After obtaining approval from the Ethics Board of the Children's Hospital of Fudan University (No. 2020-179; Shanghai, China), a retrospective review of data from patients with testicular torsion complicated by cryptorchidism was performed. Patient informed consent was waived due to the retrospective nature of the study. From January 2010 to December 2019, 22 patients were identified and treated at our tertiary pediatric medical center (Children's Hospital of Fudan University). Patients were categorized into two groups: the prepubertal group (age: 1 month to 9 years) and the postpubertal group (age: 10–16 years). Neonatal cases were excluded from the prepubertal group, and the 10-year age cutoff was based on the World Health Organization (WHO) data.4 The medical records of the patients were reviewed, and the following data were extracted: age at presentation, clinical presentations, physical examination, surgical findings, and follow-up outcomes.

The diagnosis of cryptorchid testicular torsion in children was made clinically and confirmed by emergency ultrasound and inguinal canal exploration. Surgical exploration was performed through an inguinal incision. All patients underwent immediate detorsion of the spermatic cord. Either orchiopexy or orchiectomy was performed according to the vitality of the testis. If the testis became ruddy and flexible after manual manipulation, dartos pouch orchiopexy was performed. If the testis remained dark in color after wrapping it in warm saline-soaked towels for at least 5 min, the tunica albuginea was

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Incised. If no new bleeding was identified after 15 min, orchidectomy was performed (Figure 1). Histopathological examinations were performed on all the excised testicles (Figure 2). The patients were followed at 1 week, 3 months, and 6 months after the operation and then were examined once a year. Evaluations consisted of physical examinations and ultrasonography.

The primary outcomes were the differences in the orchidectomy and salvage rates in patients with cryptorchid testicular torsion between the prepubertal and postpubertal groups. We also examined the recovery of testicular blood flow at surgery, the presentation of symptoms, the rate of misdiagnosis, and the duration of symptoms. Descriptive statistics were used to characterize clinical and management-related data. The median, interquartile range (IQR), mean, and standard deviation (s.d.) were calculated for continuous variables. Univariate analyses using Fisher's exact test for categorical variables and Student's t-test or the Mann–Whitney U test for continuous variables were used to compare variables between the prepubertal and postpubertal groups. All statistical analyses were performed with IBM SPSS Statistics for Windows, version 25.0 (IBM Corp., Armonk, NY, USA), with a two-tailed alpha of 0.05. P < 0.05 was considered statistically significant.

RESULTS

A total of 24 pediatric patients underwent surgical exploration and received a diagnosis of acute cryptorchid testicular torsion at our institution. The prevalence of UDT torsion was 13.0% (24/185) among all testicular torsion cases. Two patients were excluded because they were neonates. A total of 22 patients were included in the final analysis. The prepubertal group consisted of 12 patients (54.5%), with seven cases on the left side, and the postpubertal group consisted of 10 patients (45.5%), with nine cases on the left side. Two prepubertal patients had bilateral cryptorchidism before surgery. The median age in the prepubertal group was 23 (IQR: 6–53.25) months, while the median age in the postpubertal group was 148 (IQR: 123.75–157.25) months. The patient characteristics and the operative findings of the two groups are presented in Table 1.

The most common clinical presentations in both groups were inguinal pain, a tender inguinal mass, and an empty ipsilateral hemiscrotum. Vomiting was more prevalent in the prepubertal group (n = 5, 41.7%) than that in the postpubertal group (n = 2, 20.0%), but it was not statistically significant (P = 0.277). Restlessness at presentation was significantly more prevalent in the prepubertal group (n = 4, 33.3%) than in the postpubertal group (n = 0, 0%; P = 0.044). Although the differences were not significant, misdiagnosis was more frequent in the prepubertal group (n = 6, 50.0%) than that in the postpubertal group (n = 2, 20.0%; P = 0.145); in addition, the median duration of symptoms was longer (prepubertal: 30 [IQR: 10.75–54.50] h vs postpubertal: 9 [IQR: 5.50–40.50] h; P = 0.159), and the median degree of torsion was more severe (prepubertal: 540° [IQR: 360°–720°] vs postpubertal: 360° [IQR: 180°–810°]; P = 0.283) in the prepubertal group. After testicular torsion reduction, blood supply recovered during surgery in three of the 12 prepubertal patients (25.0%) and eight of the 10 postpubertal patients (80.0%; P = 0.010). Orchidectomy was performed in six of the 12 prepubertal children (50.0%) and two of the 10 postpubertal children (20.0%; P = 0.145). Among the six prepubertal children who underwent orchioectomy, the parents of three patients rejected orchioectomy treatment.

The mean duration of follow-up was 69.75 (s.d.: 29.62) months in the prepubertal group and 69.40 (s.d.: 25.46) months in the postpubertal group (P = 0.977). Of the 22 patients, testicular salvage was successful in two of the 12 prepubertal children (16.7%) and six of the 10 postpubertal children (60.0%; P = 0.035).

DISCUSSION

The clinical manifestations of cryptorchid testicular torsion in prepubertal children were different from those in postpubertal children according to the study data. This study also demonstrated that there was an overall low salvage rate, with a significant difference between prepubertal and postpubertal children.

Cryptorchidism, or UDT, is one of the most common pediatric abnormalities of the urogenital system, with an incidence of up to 40%–45% in preterm newborns, 1%–5% in full-term neonates at birth, 1%–2% in 3-month-old babies, and nearly 1% in 1-year-old boys. In addition to infertility and germ cell tumors, cryptorchidism is a risk factor for testicular torsion, and the risk is approximately 10 times higher than that in those with normal testicles. Testicular torsion in patients with cryptorchidism accounts for 6.8%–9.7% of all testicular torsion cases. In contrast, the incidence rate of cryptorchid testicular torsion, which mainly occurs in infancy and puberty, accounted for 13.0% of all testicular torsion cases in our series. Cryptorchidism mainly occurs in infancy and adolescence. This bimodal distribution is similar to that for all testicular torsion cases, but more cases need to be analyzed to confirm this finding.

The typical physical examination findings in patients with cryptorchid testicular torsion include a painful inguinal mass and an empty ipsilateral hemiscrotum. However, cryptorchid testicular torsion often initially manifests as atypical clinical symptoms, such as abnormal abdominal or groin pain, nausea and vomiting, restlessness, and fever, which may cause delays in medical attention seeking or result in misdiagnosis. In the present study, the data showed that restlessness was significantly more prevalent in the prepubertal group than that in the postpubertal group. Generally, due to unclear language expression, the...
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Table 1: Characteristics and surgical findings

| Variable                               | Prepuberty group (n=12) | Postpuberty group (n=10) | P    |
|----------------------------------------|-------------------------|--------------------------|------|
| Patient, n (%)                         | 12 (54.5)               | 10 (45.5)                |      |
| Age (month), median (IQR)              | 23 (6–53.25)            | 148 (123.75–157.25)     | 0.097|
| Side affected, n (%)                   |                         |                          |      |
| Right                                  | 5 (41.7)                | 1 (10.0)                 |      |
| Left                                   | 7 (58.3)                | 9 (90.0)                 |      |
| Presenting symptoms, n (%)             |                         |                          |      |
| Inguinal pain                          | 11 (91.7)               | 8 (80.0)                 | 0.427|
| Abdominal pain                         | 1 (8.3)                 | 2 (20.0)                 | 0.427|
| Vomiting                               | 5 (41.7)                | 2 (20.0)                 | 0.277|
| Restless                               | 4 (33.3)                | 0 (0)                    | 0.044|
| Fever                                  | 1 (8.3)                 | 1 (10.0)                 | 0.892|
| Limping                                | 2 (16.7)                | 0 (0)                    | 0.176|
| Scrotal swelling                       | 2 (16.7)                | 0 (0)                    | 0.176|
| Findings on physical examination, n (%)|                         |                          |      |
| Tender inguinal mass                   | 12 (100.0)              | 9 (90.0)                 | 0.262|
| Scrotal swelling                       | 2 (16.7)                | 0 (0)                    | 0.176|
| Misdiagnosis, n (%)                    | 6 (50.0)                | 2 (20.0)                 | 0.145|
| Duration of symptoms (h), median (IQR) | 30 (10.75–54.50)        | 9 (5.50–40.50)           | 0.159|
| Degree of torsion (°), median (IQR)    | 540 (360–720)           | 360 (180–810)            | 0.283|
| Ipsilateral surgical procedure, n (%)  |                         |                          |      |
| Orchiectomy                            | 6 (50.0)                | 8 (80.0)                 | 0.145|
| Orchiopexy                             | 6 (50.0)                | 2 (20.0)                 |       |
| Recovery of testicular blood flow, n (%)| 3 (25.0)                | 8 (80.0)                 | 0.010|
| Ipsilateral testicular salvage, n (%)  | 2 (16.7)                | 6 (60.0)                 | 0.035|
| Follow-up time (month), mean (s.d.)    | 69.75 (29.62)           | 69.40 (25.46)            | 0.977|

s.d.: standard deviation; IQR: interquartile range

symptoms of early-stage testicular torsion in infants and young children manifest as inconsolable crying or restlessness. Because cryptorchid torsion is a relatively rare phenomenon, physicians might not recognize cryptorchid testicular torsion, subsequently leading to misdiagnosis.

The overall rate of misdiagnosis in our patients was 36.4%; the rate in the prepubertal group was 50.0%, which was higher than that in adults (20%). Initially, abdominal pain, vomiting, and restlessness were misdiagnosed as abdominal colic or gastroenteritis in three of the prepubertal patients and misinterpreted as appendicitis in one of the postpubertal patients. One prepubertal case and one postpubertal case were misdiagnosed as posttraumatic inflammation. The clinical manifestations of inguinal masses with vomiting may easily lead to misdiagnosis as incarcerated indirect hernia, as occurred in two prepubertal patients. Inguinal hernia concomitant with cryptorchid testicular torsion is also difficult to diagnose. Visible inguinal swelling may occur as a local reaction in subcutaneous tissue caused by the ischemic/necrotic testis. Therefore, inguinal swelling is a manifestation of UDT torsion after a period of time, but not immediately.

Furthermore, necrosis of the torsioned testis led to local inflammation and subsequently inguinal erythema and fever in two of our patients. Notably, this is of little significance in differentiating UDT torsion from inguinal incarcerated hernia, so for both, emergency surgical exploration through an inguinal incision should be performed. In addition, prophylactic orchiopexy of the contralateral testis is recommended. In our study, left cryptorchidism torsion in one patient occurred 2 months after resection of the right twisted necrotic testis by a local surgeon.

The duration and the degree of spermatic cord twisting are critical factors in the outcome of testicular torsion. Although there was no significant difference between the two groups in our study, the duration of symptoms was longer and the degree of torsion was more severe in the prepubertal group than that in the postpubertal group. Moreover, the prolonged duration of symptoms in the prepubertal patients also suggests that delayed presentation and misdiagnosis are likely important factors contributing to delayed diagnosis, surgical exploration, and, subsequently, orchiectomy or salvage.

There was a poor overall rate of surgical salvage (36.4%) in our study, and the testicular salvage rate in patients with cryptorchidism can be worse, ranging from 0% to 53.8%. However, the rate of testicular salvage in the postpubertal group (60.0%) was significantly higher than that in the prepubertal group (16.7%; P = 0.035). Although the orchiopexy rate in the prepubertal group was 50.0%, the affected testis had atrophied (on physical examination and ultrasonography) in three patients whose parents rejected orchiectomy treatment. The above results may be attributed to the different clinical characteristics between the prepubertal group and the postpubertal group.

The limitations of this study include its retrospective design and the relatively small number of patients. There are differences in growth and development among children in different age groups; the age range in this study was wide, which might have biased the final results. In the future, multicenter studies with a larger number of enrolled patients to reduce bias are required. However, the number of patients in this study may be the largest number reported for cryptorchid testicular torsion in children. We hope the present study will provide important information that supplements data reported in the literature and aids physicians in diagnosing this uncommon disease.

CONCLUSION
Cryptorchid testicular torsion before puberty is different from that after puberty. Although both of the age groups underwent the same...
physical examination for a painful mass in the groin and vacuity in the scrotum, their symptoms were different. In the early stage of onset, crying and restlessness were more common in the prepubertal group than those in the postpubertal group. The salvage rate was very low in the prepubertal children. Physicians and parents should be aware of the possibility of testicular torsion in the presence of cryptorchidism, especially in prepubertal patients.

**AUTHOR CONTRIBUTIONS**
HJZ conceptualized and designed the study, acquired and analyzed the data, and drafted the manuscript. LFT participated in data acquisition and analysis and drafted the manuscript. YLB conceptualized and designed the study and data analysis. All authors read and approved the final manuscript.

**COMPETING INTERESTS**
All authors declared no competing interests.

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