The Role of Demographic and Clinical Characteristics in Distinguishing Testicular Torsion from Torsion of the Appendix Testis: A Single-center Retrospective Study

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Abstract: The acute scrotum (AS) in the pediatric population is a medical emergency. AS is usually caused by testicular torsion (TT) and torsion of the appendix testis (TAT). The current study explored which demographic and clinical characteristics can help distinguish between TT and TAT. We analyzed all children ≤16 years who underwent surgical exploration for AS. The patients were divided into Group 1/TT and Group 2/TAT. Ninety patients were included in the study (24 with TT and 66 with TAT). The peak incidence of TT was significantly higher than in the TAT group ($p<0.001$). Scrotal pain was more prevalent in the TAT group ($p=0.02$), whereas systemic signs (nausea/vomiting and abdominal pain) affected more frequently the TT patients ($p=0.003$ and $p<0.001$, respectively). The duration of symptoms was significantly longer in the TAT group ($p<0.001$). The duration of symptoms in the TT cohort significantly impacted the testicular salvage ($p=0.008$). Color Doppler ultrasound (CDUS) findings of absent/decreased testicular blood flow in the affected testis strongly favored the diagnosis of TT ($p<0.001$). The older age, shorter duration of symptoms, systemic signs, and CDUS findings can help distinguish between the two most common acute scrotum causes.

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Introduction

In childhood or adolescence, the acute scrotum (AS) is a medical emergency characterized by acute scrotal pain with or without swelling and erythema (Gatti and Murphy, 2008). The most common pathologies encountered in the broad spectrum of pediatric AS are torsion of the appendix testis (TAT), testicular torsion (TT), and epididymo-orchitis (EO). The most important differential diagnosis for AS is TT, which accounts for up to 25% of acute scrotal disease in the pediatric population (Lemini et al., 2016). Testicular torsion (Figure 1A) can occur at any age but usually occurs in young males, with a bimodal incidence in the pediatric population: during the first year of life and between the ages of 13 and 16 (Pogorelic et al., 2016).

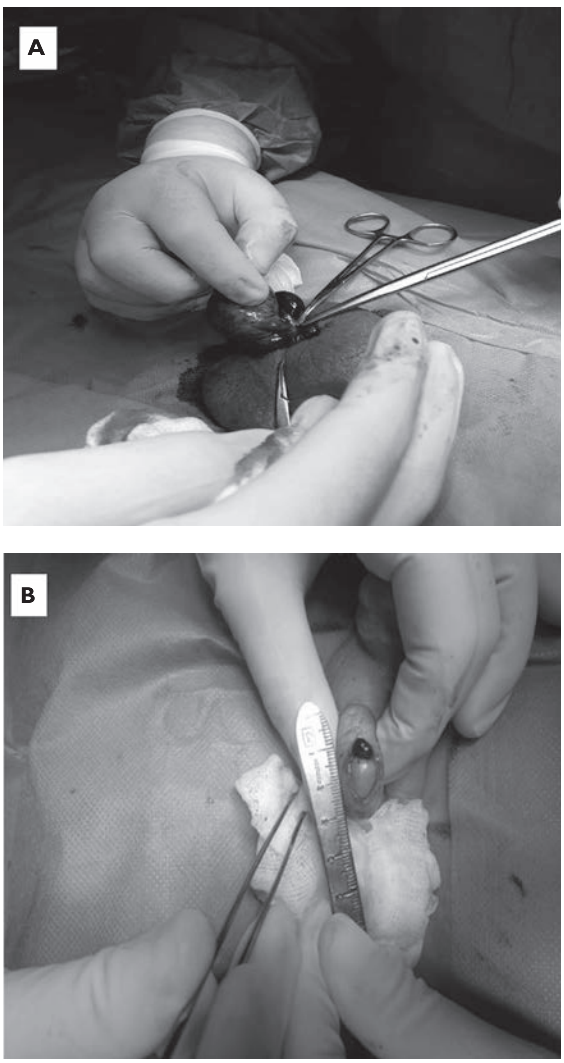

Figure 1A and B – Intraoperative images of a testicular torsion of a 12-year-old boy (A) and a torsion of the appendix testis of a 7-year-old boy (B).

In contrast, the most frequently detected pathology during scrotal exploration is TAT (Figure 1B), representing 54–71% of the operative diagnosis of AS (Mushtaq et al., 2003; Hart et al., 2016; Lala et al., 2019). TAT makes up 95% of torsed appendices (Vijayaraghavan, 2006).

Clinical distinction between TT and TAT is frequently tricky but critical because timely assessment and intervention in TT cases can preserve the affected testicle (Boettcher et al., 2012; Kumar et al., 2020). If treated within 6 hours of the presenting pain, there is a good chance of saving the affected testicle, as 90–100% of testicles will be saved. If treated within 6–12 h, depending on the degree of the torsion, 20–50% of testicles will be saved, and if treated within 12–24 h, only 0–10% of testicles will be saved (Pogorelic et al., 2021).

Various studies explored the differentiation of TT from TAT (Ciftci et al., 2004; Fujita et al., 2017; Tanaka et al., 2020). However, differential diagnosis of AS, particularly TT, remains challenging and shows a considerable risk of misdiagnosis (Ciftci et al., 2004).

The present study analyzed the pediatric cohort of TT and TAT and explored the impact of baseline demographic and clinical characteristics on differential causes of AS.

Material and Methods

We conducted a retrospective study using a cohort of pediatric patients presenting with AS who underwent scrotal exploration at the University Clinical Center Sarajevo, Bosnia and Herzegovina, between 2012 and 2016. Data were obtained from the medical records. We divided the patients into two groups according to the operative findings: Group 1 or TT group and Group 2 or TAT group and compared baseline demographic and clinical characteristics between them. The patients with TT were divided into two subgroups according to testicular viability and the type of treatment: orchidectomy and orchidopexy groups.

The primary outcome of the study was to explore the difference between testicular torsion and torsion of the appendix testicles based on their baseline demographic and clinical characteristics. The secondary outcome was determining the association between symptom duration and degree of torsion with treatment outcome (orchidectomy vs. orchidopexy) in boys and adolescents with testicular torsion.

Inclusion criteria for the study were confirmed diagnosis of TT or TAT and age below 16 years. Those with other causes of AS and those with incomplete or missing data were excluded from the study.

The following demographic and clinical characteristics were recorded: age, laterality (right/left), presenting symptoms (scrotal pain, erythema of the scrotal skin, swelling, nausea/vomiting, abdominal pain, and fever), duration of symptoms, seasonality, history of scrotal trauma, and color Doppler ultrasound (CDUS). Patients' age was categorized into five groups: <1 year, 1–3, 4–6, 7–11, and 12–16 years.

All patients underwent testicular ultrasonography with CDUS before surgery. The access for surgical exploration of the testis in all cases was through the midline scrotal incision. TT was defined as twisting the spermatic cord and its contents with resultant ischemia due to compromised blood flow to the testicle. TAT was defined as twisting and ischemia of the testicular appendage located on the superior pole of the testicle between the testis and epididymis.

All medical records were de-identified and pseudo-anonymized for the current study. The study was approved by the local institutional review board (IRB) (Ethical Committee of the Clinical Center, University of Sarajevo, 0901-2-678/18). The IRB waived informed consent due to the retrospective nature of the study.

Statistical analysis

Mean and median were used to measure central tendency, standard deviation, and range as dispersion measures for continuous variables. The values of categorical variables were presented as numbers or percentages. The Kolmogorov-Smirnov test tested the normality of data distribution for each variable. Chi-square and Fisher's exact tests were used to explore the differences between the categorical variables. P-values < 0.05 were considered significant. All statistical assays were performed using the Statistical Package for the Social Sciences (SPSS) IBM Version 26 (SPSS) (UNICOM Systems, Inc.).

Results

The baseline demographic and clinical characteristics of the two pediatric cohorts are presented in Table 1.

Ninety-eight pediatric patients with AS were identified in the period 2012–2016. Eight patients were excluded from the study as they had other pathologies (e.g., EO, n=4) or incomplete clinical histories (n=4). Thus, 90 patients with TT and TAT met the inclusion criteria and constituted the final cohort.

Twenty-four TT cases (26.6%) (Group 1) and 66 (73.4%) TAT cases (Group 2) were seen during the study period. Patients with TT in this study were significantly older (13.5 ± 2.6 years [range, ten days – 15.8 years]) than those with TAT (9.5 ± 2.8 years [range 0.7–14.7 years]) ($p<0.001$).

Although TT and TAT affected children of different ages, significant differences in both groups were observed. Thus, the peak incidence of TT was in the age of 12–16 years (75%), whereas the peak of TAT was in the age group of 7–11 years (57%) ($p<0.001$ for both calculations).

There was no statistically significant difference between the two groups in laterality ($p=0.28$). However, left-sided scrotal involvement was more common in TT cases (66%), whereas TAT cases had no significant difference in affected sides. We found that right-sided TT increases in adolescent patients: left-side TT involvement was recorded in 100% of patients under 12 years. In comparison, the incidence on that side dropped to 55.6% in patients aged ≥12.

Scrotal pain, erythema of the scrotal skin, and scrotal swelling were the most common clinical symptoms in both observed groups (Table 1). Interestingly, scrotal

Table 1 – Baseline demographic, clinical characteristics and ultrasonography findings of patients with testicular torsion and torsion of the appendix testis

Variable	Testicular torsion	Torsion of the appendix testis	P-value
Demographic characteristics of the cohort			
Patients (n=90)	24 (26.6%)	66 (73.4%)	
Age (year; median ± SD)	13.5 ± 2.6	9.5 ± 2.8*	**<0.001**
<1 year	2 (8.3%)	1 (1.5%)	**<0.001**
1–3 years	1 (4.2%)	5 (7.7%)	
4–6 years	2 (8.3%)	6 (9.2%)	
7–11 years	1 (4.2%)	37 (57%)	
12–16 years	18 (75%)	16 (24.6%)	
Laterality (n %)			0.28
Right side	8 (33.3%)	33 (50%)	
Left side	16 (66.6%)	32 (48.5%)	
Both sides	0 (0%)	1 (1.5%)	
History of trauma			0.50
Yes	2 (8.3%)	11 (16.7%)	
No	22 (91.7%)	55 (83.3%)	
Presenting clinical symptoms			
Scrotal pain (n %)	20 (83.3%)	65 (98.5%)	**0.02**
Erythema (n %)	20 (83.3%)	51 (77.3%)	0.77
Swelling (n %)	13 (54.2%)	44 (66.6%)	0.33
Nausea/vomiting (n %)	7 (29.2%)	3 (4.5%)	**0.003**
Abdominal pain (n %)	6 (25%)	0 (0%)	**<0.001**
Fever (n %)	2 (8.3%)	0 (0%)	0.07
Duration of symptoms	mean: 24 hours, median: 48 hours (range, 30 minutes – 480 hours)		
≤6 hours	9 (37.5%)	7 (10.6%)	**<0.001**
6–12 hours	0 (0%)	4 (6%)	
12–24 hours	7 (29.2%)	5 (7.5%)	
>24 hours	8 (33.3%)	50 (75.7%)	
Season			0.31
Spring	4 (16.6%)	18 (27.2%)	
Summer	8 (33.3%)	11 (16.7%)	
Autumn	7 (29.2%)	18 (27.2%)	
Winter	5 (21%)	19 (28.7%)	
Color Doppler ultrasound findings			
Absent/decreased flow	23 (96%)	5 (7.5%)	**<0.001**
Increased/normal flow	1 (4%)	61 (92.5%)	

Only significant variable values are bolded. *Age is missing for one patient in this category. SD – standard deviation

Table 2 – The duration of symptoms was significantly associated with the treatment options (p=0.008, Fisher's exact test). Similar results (77.5% vs. 37.5%) were obtained when the variable duration of symptoms was dichotomized by the median value (12 hours) (p=0.038, Fisher's exact test)

Patients with testicular torsion (n=24)		Treatment		Total
		orchidectomy	orchidopexy	
Duration of symptoms	<42 hours*	4 (25.0%)	12 (75.0%)	16 (100%)
	>42 hours	7 (87.5%)	1 (12.5%)	8 (100%)
Total		11 (46.0%)	13 (54.0%)	24 (100%)

*the mean value dichotomized the variable

pain (without recorded accurate localization of tenderness and its intensity) was statistically more present in TAT patients (p=0.02). In contrast, nausea/vomiting and abdominal pain occurred more frequently among the TT patients (p=0.003 and p<0.001, respectively). Notably, fever and abdominal pain did not affect TAT patients (Table 1).

The mean duration of symptoms for the entire cohort was 63.3 hours (range 30 minutes to 480 hours) with a median of 48 hours. The mean duration of symptoms was significantly shorter in the TT group (42 hours, range, 1–336 hours with a median of 12 hours) than in the TAT group (71 hours, range, 1–480 hours with a median of 48 hours) (p<0.001).

Among twenty-four patients with TT, 11 cases (46%) had undergone orchidectomy, and 13 cases (54%) had orchidopexy (Table 2). The duration of symptoms significantly impacted the treatment outcome (orchidectomy vs. orchidopexy, p=0.008) (Table 2).

There was no significant difference in the seasons of onset between TT patients and those with TAT (p=0.31). The lowest TT cases (21%) were recorded during the winter, whereas the largest TAT cases (28.7%) were recorded during the same season.

Most cases (91.7% of TT and 83.3% of TAT cases) had spontaneous torsion, whereas the remaining 8.3% of TT cases and 16.3% of TT cases were trauma-related. However, the difference was not statistically significant (p=0.50).

Ultrasonographically, the two diseases presented strikingly different. Thus, twenty-three patients with TT (96%) and only five patients with TAT (7.5%) showed absent or decreased testicular blood flow in the affected testes, whereas 61 (92.5%) patients with TAT and only 1 (4%) patient with TT showed increased or normal testicular blood flow in the affected testes (Table 1). CDUS findings of absent or decreased testicular blood flow in the affected testes significantly correlated with TT's presence (p<0.001). In contrast, CDUS findings of increased or normal blood flow significantly correlated with the presence of TAT (p<0.001).

Discussion

It is well documented that TAT is the most common cause of AS in children who underwent scrotal exploration (Murphy et al., 2006). Emergency scrotal exploration is the standard management means, as no other investigation can confidently exclude TT from the differential diagnosis of AS (Cavusoglu et al., 2005). Our results for the occurrence of TT and TAT are in line with the previous studies (Van Glabeke et al., 1999; McAndrew et al., 2002; Mushtaq et al., 2003; Cavusoglu et al., 2005; Murphy et al., 2006; Yang et al., 2011). Of pediatric patients with AS who underwent emergency surgery, TT is the cause of 17–72% of cases (Van Glabeke et al., 1999; Mushtaq et al., 2003; Cavusoglu et al., 2005; Yang et al., 2011). In our study, TT occurred in ~27% of cases, whereas TAT affected 73% of the AS patients. Like other studies (Ciftci et al., 2004), the patients with TT were the eldest, and their presentation was the earliest compared with the TAT patients.

Consistent with previous studies, our findings of 24 TT patients indicate that this condition occurs more frequently on the left side (Marulaiah et al., 2010; Mukendi et al., 2020). This could be anatomically related to the greater length of the left spermatic cord, which is more prone to twisting (Williamson, 1976). Data in the literature on the association between age and laterality of TT are scarce. Our finding that right-sided TT increases in adolescent patients is consistent with the study conducted by Mukendi et al. (2020). Further studies on a larger sample are necessary to determine the clinical significance of this association.

A history of nausea/vomiting and abdominal pain, as markers of a systemic response to an ischemic event in the body secondary to celiac ganglion stimulation, strongly suggest TT. However, they are absent in more than two-thirds of patients. Similar results have been reported in other studies (Hegarty et al., 2001; Srinivasan et al., 2011). Furthermore, a series of children with TT who presented with abdominal pain without testicular pain were also reported (Pogorelic et al., 2019; Wang and Mo, 2019).

The presence of only abdominal pain or other nonspecific symptoms (nausea, vomiting or urinary symptoms) may delay the diagnosis and treatment with an increased incidence of testicular loss. In our study, only 2/24 (8.3%) of patients with TT had nonspecific symptoms without testicular pain, which is in line with previous studies by Pogorelic et al. (2019) and Wang and Mo (2019). Both patients from our cohort were misdiagnosed as having abdominal diseases. Later surgical exploration revealed that both testicles had necrotized because of the ischemia time, and consequently, an orchidectomy was performed.

We believe that the higher frequency of scrotal pain in patients with TAT could be explained by the lack of registration of the exact localization of pain and the initial presentation of a larger number of patients with advanced local scrotal findings.

Our previous study showed that the duration of symptoms is the most crucial predictor of testicular salvage following TT in children (Zvizdic et al., 2021). In the present study, the duration of symptoms was significantly different between the two groups. Like another study (Fujita et al., 2017), our patients with TT had a significantly shorter time to presentation than patients with TAT. This could be explained by the more extensive symptoms caused by TT compared with TAT symptoms, leading to an earlier visit to the doctor.

In our cohort, we found that the laterality of TT to the right side increases in adolescence. We noted that more than half of adolescent boys aged ≥12 presented with right-sided TT. Documented data in the literature on the association between age and laterality of TT are very scarce. The relationship between age and laterality of TT was investigated in only one study conducted in South Africa, which included 308 patients from 10 years old and above (Mukendi et al., 2020). They found that boys aged ≥16 years are 1.5 times more likely to present with right-sided TT than those <16 years of age, though the difference was not significant (Mukendi et al., 2020). Further studies are needed to elucidate this possible association between older adolescent age and right-sided TT.

The seasonal influence on TT or TAT is contradictory in the current literature. While some studies have found a link between cold weather and TT and TAT (Lyronis et al., 2009; Molokwu et al., 2020), other studies failed to provide this evidence (Cost et al., 2011). Our study has not found a positive association between cold weather and TT.

Although most TT and TAT cases develop spontaneously, the causes of TT and TT may be trauma-related (sports or physical activity) in ~5% of cases (Ringdahl and Teague, 2006). History of trauma was present in 8.3% of TT cases and 16.7% of TAT cases. Our study did not reveal a significant difference in the history of trauma between the examined groups.

CDUS has substantially improved patients' clinical assessment with AS, determining TT's presence and the extent and reducing the unnecessary exploration rate (Lam et al., 2005; Sung et al., 2012). On sonography, the torsed testis may be enlarged and appear hypoechoic, but sometimes it can appear normal, particularly in the first few hours (Sung et al., 2012). In the evaluation of AS, CDUS has a sensitivity of ~64–100% and a specificity of 97–100%, showing reduced or absent blood flow to the testis as a highly specific finding in most TT patients (Chmelnik et al., 2010). However, false-negative and false-positive Doppler evaluations in the diagnosis of TT have been reported in the literature. Our study's data further support the excellent clinical utility of CDUS in differentiating between TT and TAT.

Due to the common practice at our institution that scrotal exploration is considered the procedure of choice for acute scrotum, a reasonably large number of scrotal explorations of the acute scrotum during the study period were operatively diagnosed as torsion of testicular appendages. Some other studies have supported this approach. According to Murphy et al. (2006), prompt surgical intervention in all patients with AS can minimize testicular loss. Surgical treatment of twisted appendages is safe, allowing accurate diagnosis and pain relief with minimal morbidity (Tanaka et al., 2020). However, as a positive consequence of the results of this study, we believe that this dogmatic attitude will be replaced with a more conservative approach to patients with TAT in our local setting.

There are several limitations to our study. First, it is a retrospective observational study; second, it comprises a smaller number of patients with TT; and third, the study was conducted in a single institution, limiting its generalizability. The present study also lacks some essential clinical data, such as a lack of precise localization of scrotal pain.

In conclusion, distinguishing TT from other acute scrotal pathology, including TAT, is crucial for timely surgical intervention and preservation of testes affected by AS. Older age, nausea/vomiting, abdominal pain, shorter duration of symptoms, and CDUS findings of absent or decreased testicular blood flow in the affected testis can distinguish between TT and TAT.

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