An unusual presentation of testicular torsion in children: a single – centre retrospective study

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Introduction: *The aim of this study was to investigate demographic and clinical characteristics and outcomes of the treatments of the patients with an unusual presentation of the testicular torsion (TT) and to clarify their peculiarities.*

Material and methods: *From January 1999 until December 2017, the case records of 149 patients who underwent surgery for TT were retrospectively reviewed. Out of that number, 25 patients were identified with unusual presentation of an acute scrotum (14 patients who presented with an abdominal pain only, and 11 who presented with testicular torsion in inguinal canal).*

Results: *The median age of all children with TT at the time of surgery was 14 years. The duration of the symptoms varied substantially and ranged from 1 hour to 120 hours with a median of 6 hours, with only 63 (42.3%) out of the 149 patients staying below the golden 6 hours. Only 2/11 (18.2%) children of the inguinal group and 5/14 (35.71%) children of the abdominal group presented within 6 hours. In the group with inguinal TT the median age was 13 years with the median duration of symptoms of 24 hours. The symptoms were mostly abdominal pain (90.9%), followed by groin pain (45.5%) and nausea (45.5%). In 6 out of 11 children, the first physical examination did not include a genital examination. In the group with abdominal pain, the a median age was 13 years, with median duration of symptoms of 17 hours. The symptoms were limited, besides the abdominal pain, to groin pain (42.8%) and nausea (50%). In 9 out of 14 children, the first physical examination did not include a genital examination. The rate of orchidectomy in the inguinal TT group was 54.5%, while in the abdominal group 57.1%.*

Conclusion: *Testicular torsion, particulary in regard to torsion in the inguinal canal or presenting dominantly with abdominal pain can be easily misdiagnosed, but needs to be recognized on time, to salvage the affected testicle. The complete physical examination, including the genital examination, needs to be performed in each male patient presenting with lower abdominal or groin pain.*

Key Words: acute scrotum, spermatic cord torsion, children, testicular torsion, abdominal pain, testicular torsion in inguinal canal, inguinal torsion

Introduction

Testicular torsion (TT) 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.[1] Considering TT and its typical presentation, the correct and fast diagnosis should be a matter of routine. The scrotal pain guides the physician´s concern clearly towards the etiological genital area. However, children may possibly present without the indicative scrotal pain and may show different inconspicuous symptoms.[1-3] The potential variable presentation is a distinctive feature of TT. An important potential presentation of children with TT, which needs to be highlighted, is the initial lower abdominal pain and/or inguinal pain pathology.[1,2,4] This occurs especially in the early stages of the condition. In that case, the pain is characterized by shifting towards the scrotal area a few hours after the onset. Before this typical pain migration, valuable time tends to be wasted by not considering TT as a possible diagnosis. Lower abdominal pain is a symptom with an extensive list of possible differential diagnosis. Thus TT can mimic abdominal conditions, like appendicitis and hence delay or even misguide the accurate diagnosis. This often results in diminished chances to salvage the affected testicle and to prevent an orchiectomy.[2,4]

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A different unusual variant of TT can occur in the setting of cryptorchidism. The incidence of TT in the setting of cryptorchidism is approximately 10 times higher.[5] The exact mechanism behind torsion in the inguinal region is not completely understood. The relatively broad testicle in comparison to its mesentery might be a crucial factor.[6] The most common age for inguinal TT is around the age of 1.[7-9] TT in the inguinal canal can have a different symptomatic, compared to the typical TT and might resemble some abdominal pathologies.[7-9] TT can also occur on both sides and hence carries a high risk of hypogonadism.[1,7-9,10] In the case of a suspected bilateral perinatal TT, an early bilateral exploration is crucial.[10] Immediate surgical exploration is always indicated in the case of a TT, regardless of the location of the testicle.[1,9-11]

The aim of this study is to identify and highlight unusual presentations of the TT and to clarify their peculiarities, by showing demographic and clinical characteristics of patients, who were operated for TT. The focus lies on two studied groups, one including patients with only abdominal pain as the presenting symptom, while the other is composed of patients with TT in the inguinal canal. In both cases, it is a rather difficult diagnosis, because of the misleading and rare symptoms which are considered unusual.

Materials and methods

Patients

A retrospective database analysis of medical records on 149 patients, with the median age of 14 years, who underwent emergency surgery because of TT between January 1990 and January 2018 was performed. The medical records included initial medical history, anamnestic data (age, duration of symptoms before surgery, presenting symptoms), physical examination findings (side of involved testis, presence of erythema, swelling, tenderness over the testis and epididymis), localization of pain, time of hospital admission, surgical findings, degree of torsion, operating results, complications and long-term consequences.

Inclusion criteria were male patients, 0-17 years of age presented with abdominal pain as the only symptom of TT or patients presented with TT in inguinal canal. Exclusion criteria were patients older than 17 years of age, patients with typical presentation of TT, patients operated in another institutions, and followed up at our outpatient clinic and those with incomplete data, or that have been followed up less than 3 months. Regarding inclusion criteria the patients were divided into two distinct groups. The set criteria for first group (n = 14) was the presentation with abdominal pain and without the classical, pinpointing towards TT, scrotal pain. The second group (n = 11) includes all patients presented with TT in the inguinal region.

Surgery

After confirmation of TT the arrangements for the emergency exploratory surgery were immediately initiated and carried out. The affected hemiscrotum or groin in case of TT in inguinal canal, was incised horizontally and explorated. The next step was an instant manual detorsion. If the testicle showed signs of viability, an orchiopexy was subsequently performed. In the case of a shady and darker appearance of the testis, the testicle was enveloped into warm saline-soaked compresses, for a minimum of 5 minutes. Possible changes of the color of the testicle were observed. If no improvement was detectable, the surgeon performed an incision into the tunica albuginea of the testis, to check for further signs of possible viability. The absence of bright bleeding was considered as an indicator to chose an orchiectomy. The removed testicles were sent to the pathology department, where the necrotic state of the organ was confirmed.

Follow up

Regular checkups after the surgery were made in the time span of 7, 30, 90 and 365 days and subsequently once a year for detection of testicular atrophy. Additional to the exploratory surgery, a few months following the operation, an implantation of a silicone prosthesis was carried out in most of the cases.

Results

The duration of the symptoms varied substantially and ranged from 1 hour to 120 hours with a median of 6 hours, with only 63 out of 149 patients staying below the golden 6 hours. The symptoms of the 149 patients, who underwent surgery for TT, varied from pain and tenderness to groin pain and elevated temperature. The most common presenting clinical symptoms were scrotal edema, scrotal pain/tenderness and an absent cremasteric reflex with an occurrence of around 87%, 85% and 82%, respectively, Table 1. Abdominal pain, groin pain and elevated temperature (> 37°C) represent with 19%, 7% and 5%, the least common symptoms. The symptoms distribution in the inguinal TT group shows clearly, that in those 11 children abdominal pain (90.9%) predominated, followed by groin pain (45.45%) and nausea (45.45%). In the abdominal pain group, evidently abdominal pain (100%) is the

TABLE 1. **Characteristics, operative findings and outcome of all patients with testicular torsion**

Patient characteristics	Testicular torsion (n = 149)	Inguinal torsion (n = 11)	Abdominal pain (n = 14)
Demographic data			
Age, median (range)	14 (0-17)	13 (0-16)	13 (3-17)
Lateralization			
Left, n (%)	84 (56)	5 (45.45)	5 (36)
Right, n (%)	65 (44)	6 (54.54)	9 (64)
Clinical data			
Symptom duration (h), median (range)	6 (1-120)	24 (3-60)	17 (3-72)
< 6 h, n (%)	63 (42.28)	2 (18.18)	5 (35.71)
6-12 h	35 (23.48)	2 (18.18)	
12-24 h	18 (12.08)	2 (18.18)	5 (35.71)
24-72 h	26 (17.44)	5 (45.45)	4 (28.57)
> 72 h	7 (4.69)		
Scrotal pain, n (%)	127 (85.23)		
Scrotal edema, n (%)	129 (86.58)	1 (9.09)	
Scrotal erythema, n (%)	78 (52.35)	1 (9.09)	
Nausea, n (%)	29 (19.46)	5 (45.45)	7 (50)
Groin pain, n (%)	17 (11.4)	5 (45.45)	6 (42.83)
Absent cremasteric reflex, n (%)	122 (81.88)		
Hard testis, n (%)	83 (55.7)		
Horizontal testice, n (%)	94 (63.09)		
Abdominal pain, n (%)	28 (18.79)	10 (90.9)	14 (100)
Torsion degree, median (range)	360 (90-1080)	360 (180-720)	360 (180-720)
Treatment outcomes			
Operational procedure			
Detorsion and fixation, n (%)	104 (69.8)	5 (45.45)	6 (42.86)
Orchidectomy, n (%)	45 (30.2)	6 (54.54)	8 (57.14)
Complications, n (%)	2 (1.34)		1 (7.14)

symptom of focus and was accompanied by groin pain (42.8%) and nausea (50%) in around half of the cases. The demographic data, clinical data and treatment outcome in all patients with TT, as well as in patients with an inguinal torsion and in those presenting with abdominal pain only are summarized in Table 1. In the majority of the cases (69.8%) a detorsion and fixation was performed, whereas in 45/149 (30.2%) patients an orchidectomy was necessary.

TT in inguinal canal

A more extensive list about the characteristic findings about the group of the 11 patients with the inguinal TT is shown in Table 2. The age distribution ranged from 0 to 16 years with a median age of 13 years. The median duration of symptoms in the inguinal group was 24 hours (range 3-60 hours). In 2/11 (18.2%) cases the patients presented within 6 hours. The symptoms were mostly limited to abdominal pain and groin pain, but also scrotal erythema, edema and nausea was reported. In 6 out of 11 children, the first physical examination did not include a genital examination. In five cases, where the patients directly received a genital examination, the first examination was carried out four times by physicians, who are specialized in the pediatric field and one time by general practitioners. After the referral in all cases to whether a pediatrician or a pediatric surgeon, the

Patient	Age	Side	Duration of symptoms (h)	Presenting symptoms	Genital examination				Intra-operative findings	Degree of TT	Outcome
					1st examination		2nd examination				
					Genital examination	Testicular torsion Recognized – YES/NO	Genital examination	Testicular torsion Recognized – YES/NO			
1	14	L	56	Groin pain, scrotal edema, erythema, nausea	NO	NO (ER physician)	YES	YES (pedaitric surgeon)	Testicular gangrene	360°	Orchidectomy
2	13	R	5	Abdominal pain, groin pain	YES	YES (pediatrician)	-	-	Inguinal torsion	720°	Salvaged
3	15	L	60	Abdominal pain, nausea	NO	NO (pediatrician)	YES	YES (pedaitric surgeon)	Testicular gangrene	360°	Orchidectomy
4	16	R	4	Abdominal pain	YES	YES (ER physician)	-	-	Testicular gangrene	360°	Salvaged
5	15	R	6	Abdominal pain, groin pain, nausea	YES	YES (pediatric surgeon)	-	-	Testicular torsion	270°	Salvaged
6	1	L	36	Abdominal pain	NO	NO (ER physician)	YES	YES (pediatrician)	Testicular gangrene	360°	Orchidectomy
7	0	R	16	Abdominal pain, groin pain, nausea	YES	YES (pediatrician)	-	-	Testicular gangrene	180°	Salvaged
8	9	R	48	Abdominal pain	NO	NO (ER physician)	YES	YES (pediatrician)	Testicular gangrene	360°	Orchidectomy
9	13	L	3	Abdominal pain	YES	YES (pediatrician)	-	-	Testicular torsion	360°	Salvaged
10	1	L	48	Abdominal pain, nausea	NO	NO (pediatrician)	YES	YES (pediatrician)	Testicular gangrene	360°	Orchidectomy
11	0	L	24	Abdominal pain, groin pain	NO	NO (ER physician)	YES	YES (pediatric surgeon)	Testicular gangrene	360°	Orchidectomy

Table 2. Patient characteristics, operative findings and outcome in group of patients presenting with torsion in the inguinal canal (n = 11).

Patient	Age	Side	Duration of symptoms (h)	Presenting symptoms	Genital examination				Intraoperative findings	Degree of TT	Outcome
					1st examination		2nd examination				
					Genital examination	Testicular torsion Recognized – YES/NO	Genital examination	Testicular torsion Recognized – YES/NO			
1	14	R	3	Abdominal pain, nausea	YES	YES (pediatrician)	-	-	Testicular torsion	720°	Salvaged
2	13	R	5	Abdominal pain, groin pain	NO	NO (ER physician)	YES	YES (pediatric surgeon)	Inguinal torsion	720°	Salvaged
3	9	L	18	Abdominal pain, nausea	NO	NO (GP)	YES	YES (pediatric surgeon)	Testicular gangrene	360°	Orchidectomy
4	16	L	36	Abdominal pain	NO	NO (GP)	YES	YES (pediatrician)	Testicular gangrene	360°	Orchidectomy
5	12	R	3	Abdominal pain, groin pain, nausea	YES	YES (pediatrician)	-	-	Testicular torsion	360°	Salvaged
6	13	L	24	Abdominal pain	NO	NO (resident of surgery)	YES	YES (pediatric surgeon)	Testicular gangrene	360°	Orchidectomy
7	16	R	48	Abdominal pain, groin pain, nausea	NO	NO (ER physician)	YES	YES (pediatric surgeon)	Testicular gangrene	270°	Orchidectomy
8	3	R	72	Abdominal pain	NO	NO (pediatrician)	YES	YES (pediatric surgeon)	Testicular gangrene	720°	Orchidectomy
9	17	L	4	Abdominal pain	YES	YES (GP)	-	-	Testicular torsion	360°	Salvaged
10	17	R	16	Abdominal pain, nausea	NO	NO (pediatrician)	YES	YES (pediatrician)	Testicular gangrene	540°	Orchidectomy
11	12	R	10	Abdominal pain, groin pain	YES	YES (pediatric surgeon)	-	-	Testicular gangrene	180°	Salvaged
12	13	R	5	Abdominal pain	YES	YES (GP)	-	-	Testicular torsion	180°	Salvaged
13	12	R	18	Abdominal pain, groin pain, nausea	NO	NO (ER physician)	YES	YES (pediatric surgeon)	Testicular gangrene	360°	Orchidectomy
14	12	L	19	Abdominal pain, groin pain, nausea	NO	NO (GP)	YES	YES (pediatric surgeon)	Testicular gangrene	720°	Orchidectomy

Table 3. Patient characteristics, operative findings and outcome in group of patients presenting with abdominal pain (n = 14).

second physical examination always included a genital evaluation. In the second evaluation the condition of TT in the inguinal canal was recognized in all six cases. In 6/11 (54.5%) cases an orchidectomy was performed, because testicular gangrene was found intraoperatively. The follow up showed a successful outcome and no complications. There were no cases of testicular atrophy after detorsion and fixation of the testicle. All children with detorsed and fixed testicle reported no pain or any other noticeable problems.

TT presented with abdominal pain

Table 3 shows 14 selected patients out of the 149 children with TT. The main symptom and also including criteria for this group, was the abdominal pain, often combined with groin pain and vomiting, but no classical TT signs, like scrotal edema and scrotal tenderness. The age was in the range of 3-17 years with a median of 13 years. In 9 out of 14 children, the first physical examination did not include a genital examination. In those five cases, where the patients directly received a genital examination, the first examination was carried out three times by physicians, who are specialized in the pediatric field and two times by general practitioners. After the referral in all cases to whether a pediatrician or a pediatric surgeon, the second physical examination always included a genital evaluation. In the second evaluation, the condition of TT was recognized in all nine cases. The median duration of the symptoms in the observed group was 17 hours (range 3-72 hours). In 5/14 (35.7%) cases the children presented within 6 hours. Out of these 14 operated boys, the testicles of six (42.9%) were salvaged and in eight (57.1%) cases, an orchidectomy was performed because of gangrene of the testis. The follow up showed a successful outcome and no complications, except one reported wound infection. There were no cases of testicular atrophy after detorsion and fixation of the testicle. All children with detorsed and fixed testicle reported no pain or any other noticeable problems.

Discussion

TT is a pediatric emergency that most commonly occurs during puberty, but may be seen at any age. The treatment consists of an attempt to surgically explore the area and reestablish the inhibited blood flow.[1,12] A salvage attempt within 6 hours has a success rate of 90%-100%, strongly successively decreasing after that time frame. Accordingly to 25%-65% within 6-12 hours and to 0%-24% within 12-24 hours.[1,3,7] Thus the outcome is time depended. There are two essential time frames, where valuable time tends to get lost. The first one is the duration of the symptoms, until the child indicates the problem and manages to present to the emergency room. The second window, is the time the responsible physician needs to make the accurate diagnosis and initiate the treatment. Atrophy of the testicle is an unpreventable event, if the pain lasted more than 8 hours.[1,12] In general, approximately 32% of the children with TT receive an orchidectomy.[1,2,12] In 104 (69.8%) out of the 149 (100%) patients in our study, the surgeons were able to salvage the affected testicle. The duration of the symptoms ranged from 1-120 hours and 63 (42.3%) patients presented within 6 hours.

Torsion of the testicle in the inguinal canal is a rare event and infrequently reported. Candocia et al mention in their case report, the increased incidence of TT, in the setting of boys with undescended testis in the association with spastic neuromuscular disorders.[9] Cryptorchidism can be subdivided into inguinal, subinguinal or abdominal, dependent on the position of the testicle. The testis is found in 75% within the inguinal canal. Nonetheless, TT in the inguinal canal itself is quite unusual.[7,10,13] The phenomenon of an undescended testis is present in 3.5% of full-term infants, but decreases to 1% at the age of 1 year, because of its self-resolving and often just delayed nature.[7,9] Following those data, TT in the inguinal canal is expected to peak at the age of 1 year, even though the mean age in our study is 8.81 years. A possible explanation is, that the condition of cryptorchidism was not recognized before, because those patients were mostly from rural areas and did not visit any medical institution. The case of a 14-month-old boy, who refused weight bearing on his left leg, turned out to be a testicular torsion in the inguinal canal. This report exemplifies the possible obscurity of that condition.[14] The symptoms of an inguinal torsion can resemble an appendicitis or other similar conditions, connected to the umbrella term – acute abdomen.[7,13-15] The goal of the treatment of torsion in the inguinal canal is a testicular preserving orchiopexy, if the testis is still viable. In a study, carried out by Zilberman et al, 5/11 boys received an orchidectomy.[15] In general, the few available studies and case reports containing the topic of TT and undescended testis state, that the correct diagnosis is frequently delayed and the resultant orchidectomy often necessary.[7,9,16] In our study, 11 (7.4%) children, out of the total number of patients, had an inguinal TT. In 6/11 children with inguinal TT, the outcome was an orchidectomy. The mean duration of symptoms in the inguinal group was 27.8 hours. Only two patients presented within the first 6 hours. Our data show that in 6/11 cases in the inguinal

group, the first physical examination did not include a genital examination and thus the condition kept being undetected, until they were examined again. In most of the cases the first investigating physician was not a specialized pediatrician. Therefore the neglected genital examination could be a consequence of the inexperience of the physician. The second evaluation always included the genital examination and also always correctly diagnosed TT. Thus the genital evaluation represents a reliable method to diagnose TT.

Abdominal pain is a demanding symptom, with a broad spectrum of differential diagnosis.[17] The possibilities range from a simple harmless condition to an acute state of emergency. Drawing the wrong conclusion might lead to serious consequences for the patient. Hence the physician, responsible for the first examination, is obligated to investigate the pathology behind the abdominal pain by any means. Performing a complete physical examination also implies an inspection of the genital area.[3,4,18] The typical presentation of TT includes sudden onset of sharp testicular pain and scrotal swelling, which can be accompanied by additional features. The testicular innervation originates from Th 10 and Th 11. On the other hand, the anterior part of the scrotal area is innervated by L1 and the posterior one by S2/S3. So the anatomy can lead to various diffuse and misleading pain stimuli that are sensed by the patient outside of the affected area.[19] In the reported case of a boy with abdominal pain, the physician did not perform a genital examination. In a second examination a pediatrician detected scrotal bruising and a tender testis. A surgery was immediately initiated, but the testis was already gangrenous and had to be removed.[18] In another case of two boys, with abdominal pain as the leading the symptom, the genital evaluation was neglected as well. In both patients the intraoperative findings showed a non-viable testis and they had to undergo an orchidectomy.[19] A further example for the easily misdiagnosed TT, presenting with abdominal pain, is the report of a boy, who got wrongly diagnosed with appendicitis. Additional evaluations revealed an incomplete TT.[20] Anderson et al stated that 22% of their patients were troubled by abdominal pain, which frequently overshadowed the pain in the scrotal area. They also reported three cases of unnecessary appendectomies, due to the misconception of appendicitis, instead of the actual existing TT.[21] In New York an insurance company claimed, that TT is placed on the 4th position of common misdiagnosed conditions.[22] TT is also the 3rd most common reason of accusations for medical malpractice against physicians.[18] In our study 11 patients with inguinal TT and 14 patients with TT, presenting with abdominal pain, represent 16.77% of the reviewed 149 patients in total. The mean duration of the symptoms in the observed abdominal group was 20.07 hours, and 5/14 presented within the valuable 6 hours. In the abdominal pain group 8/14 children received an orchidectomy. Our results show, that no immediate genital examination was performed in 9 out of 14 cases, in the patients with abdominal pain, which possibly is due to the unfamiliar presentation.

The main limitation of this study is its retrospective character. The medical records are prone to bias. Duration of the symptoms is a subjective information and can have significant inaccuracies. Statements from children depending on the age are disputably reliable. Thus it is possible that sometimes less symptoms were reported by the patients, than they actually had.

Conclusion

The inferior outcome of the TT with an unusual presentation, reflects the difficulties of the unacquainted characteristics of these uncommon cases. The possible misleading presentation, the fact that children often have difficulties to properly express themselves, the narrow golden time window to resolve the situation – the conglomeration of those circumstances makes TT prone to false decisions and subsequent malpractice. The quintessence is to include the genital evaluation in the first physical examination in male children. The responsible physician simply needs the knowledge about the possible unusual presentations of TT, thus he can act accordingly. Announcing and emphasizing informations about the different odd presentations of TT into the common medical knowledge, could improve the general outcome. □

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