

Original Article

Perinatal Testicular Torsions: 21 Years Clinical Experience

T TARTAR, M SARAC, U BAKAL, E GENÇ, MR ONUR, A KAZEZ

Abstract

Purpose: Perinatal testicular torsion (PTT) diagnosis and treatment management are difficult due to its rare occurrence and uncertain aetiology. **Methods:** The demographic data, complaints at admission, radiologic characteristics, diagnosis and treatment methods of patients who underwent follow-up and treatment for PTT were recorded. **Findings:** Of the patients, 50% had PTT on the right testicle, 30% had PTT on left testicle, and 20% had bilateral PTT. All patients except for one had discolouration in the scrotum. During the examinations of the patients, twisted testicles were palpated as being harder than normal. In 70% of the cases, blood flow could not be visualised in colour doppler ultrasonography (CDUS). Hydrocele was present on the opposite side of the affected testicle in 60% of the cases. Orchiectomy+fixation on the opposite testicle were performed on 10 of the 12 twisted testicles. **Conclusions:** Physical examination and CDUS are the primary methods used for diagnosis of PTT. The presence of hydrocele in the opposite scrotum can be a warning with regards to PTT. Due to the potential of contralateral torsion, fixation must be performed on the opposite testicle during the same session/intervention.

Key words

Fixation; Intrauterine; Newborn; Prenatal; Testicular torsion

Introduction

Perinatal testicular torsion (PTT) is a clinical condition observed between the intrauterine period and the first 30 days of life. Clinical diagnosis and treatment management are very difficult because PTT is a rare condition with an

unknown aetiology. The prevalence of neonatal testicular torsion (TT) is 6.1/100,000, and it accounts for 12% of all TT cases.^{1,2} Of the TTs that occur during the neonatal period, 72% are observed in the intrauterine period.³ These TTs are generally unilateral.⁴ TTs often occur extravaginally and lead to infertility and endocrinological dysfunction following testicular ischaemia and atrophy.^{5,6}

In this study, we evaluated the diagnosis and treatment management of patients treated for PTT.

Department of Pediatric Surgery, Firat University Faculty of Medicine, 23119, Elazığ / Turkey

T TARTAR MD
M SARAC MD
U BAKAL MD
E GENÇ MD
A KAZEZ MD

Department of Radiology, Hacettepe University Faculty of Medicine, 06100, Ankara / Turkey

MR ONUR MD

Correspondence to: Dr T TARTAR
Email: tugaytartar@gmail.com

Received July 20 2020

Methods

Patients

Prior to the study, approval was obtained from the Non-Interventional Research Ethics Committee of Firat University (meeting date: 13/06/2019, number of meetings: 10, decision no: 06), and the study was performed in accordance with the principles of the Helsinki Declaration. A retrospective examination was performed on the records of patients aged 0-16 years who had been treated for the diagnosis of TT in the paediatric

surgery clinic of a university hospital between January 1998 and April 2019. For all the patients who were followed-up and treated for PTT, the following information was recorded in the PTT registration form: the type of delivery, gestational age, the age of diagnosis, birth weight, maternal age and number of births, complaints at admission to the hospital, physical examination findings, torsion, direction and degree, radiological characteristics, developing complications, diagnosis and treatment methods, histopathological findings, the duration of hospitalisations, follow-up periods and additional pathologies identified in the mother and the infant.

Statistical Analysis

IBM SPSS Statistics version 22 package software (SPSS Inc., Chicago, IL, USA) was used for statistical analysis. The normality of quantitative data was examined using the Shapiro–Wilk test. To define baseline demographic data and clinical characteristics, descriptive statistics, such as frequency and percentage, were used for categorical variables, whereas mean (\pm standard deviation) and median (min-max) were used for continuous variables.

Results

Perinatal testicular torsion was diagnosed in a total of 12 testes in 10 (21.7%) of 46 patients who underwent surgery for TT over a 21-year period. The median age of diagnosis was one day (min: 1, max: 12). PTT was in the right testis in five (50%) of the patients, in the left testis in three (30%), and bilateral in two (20%). Median gestational age at delivery was 38 weeks (min: 35, max: 40). Of all the 10 patients, six (60%) and four (40%) were delivered by caesarean section and vaginal delivery, respectively. Median birth weight was 3300 grams (min: 1730, max: 4000). Reasons for the admission of the infants included colour change in the scrotum, redness, swelling and small testes (Figure 1). During physical examination, the testes with torsion were palpated as painless and rigid structures. In one patient who was followed-up without surgery, the only complaint was a small and rigid testis upon palpation. One patient diagnosed with PTT on the 12th day exhibited sensitivity in the scrotum. Colour doppler ultrasonography (CDUS) was performed in all the patients. Hydrocele was identified in the contralateral scrotum of four (50%) patients with unilateral PTT and both patients with bilateral PTT. In seven (87.5%) patients with unilateral PTT, orchietomy was performed due to the

necrotic appearance of the testes and the absence of bleeding when an incision was performed on the testes after detorsion (Figure 2). For one patient (12.5%), follow-up was conducted without orchietomy due to the insistence of the family. On CDUS, this patient was diagnosed with atrophic testis. Bilateral orchietomy was performed in one of the two patients with bilateral PTT. Right orchietomy was performed on the other patient, and detorsion + fixation was performed due to the ischaemic appearance and the pink colouration of the testis after detorsion of the left testis. During the 9-month follow-up of this patient, scrotal CDUS showed a decrease in the size of the left testis (8×5×4 mm, atrophy) and a distinct heterogeneous appearance and calcifications in the parenchyma. Contralateral testicular fixation was performed in seven (87.5%) patients who had unilateral PTT and underwent surgery. The median torsion degree was 540 (min: 360, max: 720). Seven (70%) patients had clockwise torsion, whereas three (30%) had counter clockwise torsion. Nine (90%) patients had extravaginal torsion, whereas one (10%) had intravaginal torsion. Postoperative complications were not observed. Histopathological examination was performed on the material extracted from all the patients who underwent orchietomy (Table 1). The median duration of hospitalisation was 4 days (min: 2, max: 18). Median maternal age was 30 years (min: 26, max: 45). The median number of pregnancies was 3 (min: 1, max: 10). One mother had diabetes mellitus as a concomitant disease. One patient with PTT had patent ductus arteriosus, and another had hydronephrosis. The mean follow-up period was of 2 years. The demographic characteristics, complaints and radiological and histopathological findings of the patients are provided in Table 1.



Figure 1 Colour change, redness and swelling in the right scrotum.

Discussion

Testicular torsions are more common in children during the neonatal and puberty periods.⁷ Neonatal TTs are mostly extravaginal (92%).^{5,8} In our study, extravaginal PTT was detected in 90% of the patients, which is in accordance with that in the literature.

Concerning its aetiology, the prevailing view is that although the testis descends into the scrotum, TTs occur due to incomplete fixation of the tunica vaginalis.⁹ Vaginal delivery, prolonged delivery time, twin pregnancy, preeclampsia, gestational diabetes and high birth weight have been reported to be risk factors of PTT. It has been reported that 90% of infants with PTT are vaginally delivered.¹⁰ PTT is usually seen in term infants.¹¹ In accordance with the literature, nine (90%) of the patients in our study were term infants. Eight (80%) patients had a birth weight over 3000 grams. However, contrary to the literature, six (60%) and four (40%) of the patients in our study were delivered by caesarean section and vaginal delivery, respectively. We think that this difference may be due to high caesarean delivery rates in Turkey. There were no twin pregnancies or a history of preeclampsia in this study; however, as mentioned before, one of our patients was the infant of a diabetic mother.

Perinatal testicular torsion is usually asymptomatic.² Approximately 70% of the patients are diagnosed at birth.¹²



Figure 2 Necrotic left testis with, no bleeding in the incision made after detorsion.

Anamnesis, physical examination and scrotal CDUS are important for the diagnosis of PTT.⁷ In infants born with PTT, it is observed that the scrotum on the affected side is usually of a dark colour, the testis is painless and in the form of a rigid mass, the skin is attached to the mass and the internal structure cannot be differentiated. In cases of TT that occurs during or after birth, skin hyperaemia and tenderness may be detected in addition to the history of normal scrotum at birth that necessitates emergency exploration. The presence of a purple colouration in the scrotum and the palpation of rigid testis tissue are particularly important physical examination findings in the diagnosis of TT.¹³ Hydrocele is often seen on the opposite side in intrauterine TTs.¹⁴ In our study, discolouration, redness and swelling were present in the scrotum of all but one patient who was not operated. In all of the patients, the testis was palpated as a rigid tissue. PTT diagnosis was confirmed by scrotal CDUS in all the patients. Hydrocele was present in six (60%) of the patients. In accordance with the literature, six (60%) of the patients were diagnosed at birth.

Perinatal testicular torsions have been reported to be more common on the left side.^{7-9,14} In addition, 11-22% can be bilateral.⁴ Bilateral PTTs may present with symptoms at the same time or at different times.^{3,14} However, the prevailing view is that bilateral PTTs often present symptoms at the same time.¹¹ Contrary to the literature, five (50%) of the patients in our study has PTTs on the right side. In contrast, the incidence of bilateral PTT in our study was consistent with that in the literature. Bilateral PTTs had presented with symptoms at the same time.

To our knowledge, there is no data in the literature on the relationship of PTTs with gestational age and the number of pregnancies. We believe that large-scale studies are needed on this subject.

In PTTs, successful recovery of the testis is achieved in 0-21.7% of the patients.^{8,15} Moreover, torsion is seen in the contralateral testis at the rate of 5-30%.¹⁰ Even if TTs occur during the intrauterine period, testis can be recovered from torsions that occur near or during the delivery.^{3,10,14} In an experimental study, it was reported that the duration and degree of torsion were the most important factors in the development of ischaemia and necrosis.¹⁶ In the study conducted by Arda et al, instant bleeding after incision to the tunica vaginalis during surgery was classified as grade 1, bleeding within 10 minutes after the incision was classified as grade 2 and the absence of bleeding after 10 minutes was classified as grade 3. The sensitivity and specificity in the diagnosis/determination of testicular

Table 1 Demographic characteristics, complaints, torsion side and degree and radiological and histopathological

Patient	Age at Diagnosis (days)	Type of Delivery	GW	BW (g)	Complaint	Torsion Side	Torsion Degree	Radiological Findings	Histopathological Findings
1	12	C/S	35	1730	Redness, swelling and pain in the left scrotum	Left	540	Hypochoic left testis parenchyma and calcified wall	Diffuse coagulation necrosis, calcification
2	1	VD	38	3600	Discolouration in the right scrotum	Right	360	Atrophic appearance of right testis with oedematous and necrotic areas, absence of blood flow in the testis increased right epididymal size, bilateral hydrocele	Diffuse coagulation necrosis
3	5	VD	38	3200	Discolouration and swelling in the left scrotum	Left	720	Increased left testis and epididymis size, increased echogenicity in the left testis with blood flow, heterogeneous and hypochoic appearance in the epididymis, right hydrocele	Diffuse coagulation necrosis, calcification
4	1	VD	38	2500	Redness and swelling in the left scrotum	Left	360	Decreased echogenicity in the left testis with absence of blood flow, right hydrocele	Haemorrhagic, necrotic testicular tissue
5	1	C/S	40	3050	Discolouration in the right scrotum	Right	720	Increased right testis size with heterogeneous and hypochoic appearance of testicular parenchyma, increased right epididymis size with hypochoic appearance and no blood flow	Haemorrhagic, necrotic testicular tissue
6	4	C/S	38	3300	Discolouration and swelling in both scrotum	Bilateral	540	Decreased right testis size, peripheral parenchymal calcifications and increased echogenicity, increased left testis and epididymis size, no blood flow, left hydrocele	Ischaemic necrosis findings, congestion, increased fibrous tissue
7	1	C/S	38	3600	Discolouration in the right scrotum	Right	360	Increased right testis size, hypochoic testis parenchyma with no blood flow	Ischaemic necrosis findings
8	1	VD	38	3450	Discolouration and swelling in both scrotum	Bilateral	720	Heterogeneous and hypochoic appearance of bilateral testicular parenchyma with absence of blood flow, bilateral hydrocele	Haemorrhagic infarction
9	1	C/S	38	4000	Redness in the right scrotum	Right	360	Increased right testis size, heterogeneous - hypoechoic areas in the parenchyma, bilateral hydrocele	Haemorrhagic infarction
10	5	C/S	36	3100	Small right testis	Right	–	Decreased right testis size, heterogeneous - hypoechoic areas in the parenchyma with calcification	–

GW: Gestational week, BW: Birth weight, C/S: Caesarean section, VD: Vaginal delivery

viability in the study were reported as 100% and 78%, respectively, in which grade 1 and 2 patients underwent fixation, whereas grade 3 patients underwent orchiectomy.¹⁷ In our study, orchiectomy was performed in the necrotic right testis of one patient with bilateral PTT, whereas detorsion + fixation was applied in the grade 2 left testis of another patient. Orchiectomy was performed in the other patients because all other PTTs were classified as grade 3. Of the 12 testes with PTT, only one (8.3%) that was classified as grade 2 could be saved by applying detorsion + fixation.

To our knowledge, there is no consensus in the literature regarding the application of testicular fixation on the testis without torsion in extravaginal TTs.¹⁸ Surveys conducted in different clinics revealed that delayed orchiopexy, close follow-up and orchiopexy under emergency conditions were performed at rates of 57%, 33%, and 10%, respectively.¹⁹ In our study, considering the risk of anaesthesia, fixation was performed on the contralateral testis under emergency conditions in all the patients who underwent surgery. It is very difficult to recover the testis in PTTs. However, we believe that urgent intervention in PTT is necessary due to the uncertainty in the time period between the testicular torsion and patient

presentation, need for confirmation of the PTT diagnosis and evaluation of the contralateral testis.

Differential diagnosis of PTT includes incarcerated scrotal hernia, scrotal haematoma, scrotal abscess, peritonitis due to meconium, epididymitis and neoplasm.^{5,13} Differentiation PPT from mimicking diseases necessitates a detailed perinatal history, careful physical examination, adequate laboratory tests and imaging assessment. CDUS remains as the mainstay imaging technique in differentiation of PTT from abovementioned mimicking disorders. A gray-scale US assessment with its real-time imaging capability is helpful in the diagnosis of scrotal hernia, scrotal haematoma, and scrotal abscess. Meconium peritonitis may present with scrotal fluid with no finding of deterioration in testicular vascularity on CDUS. Epididymitis manifests as an enlarged epididymis with heterogeneous echotexture and increased vascularity on CDUS. Neoplasm of the testis in newborns may be easily detected on CDUS as a mass with focal or diffuse involvement in testicular parenchyma with variable vascularity. PTT may be differentiated from testis neoplasms by absence of tumour markers and with its more heterogeneous appearance on gray-scale US and diminished or absence of vascularity in the testis (Figure 3).

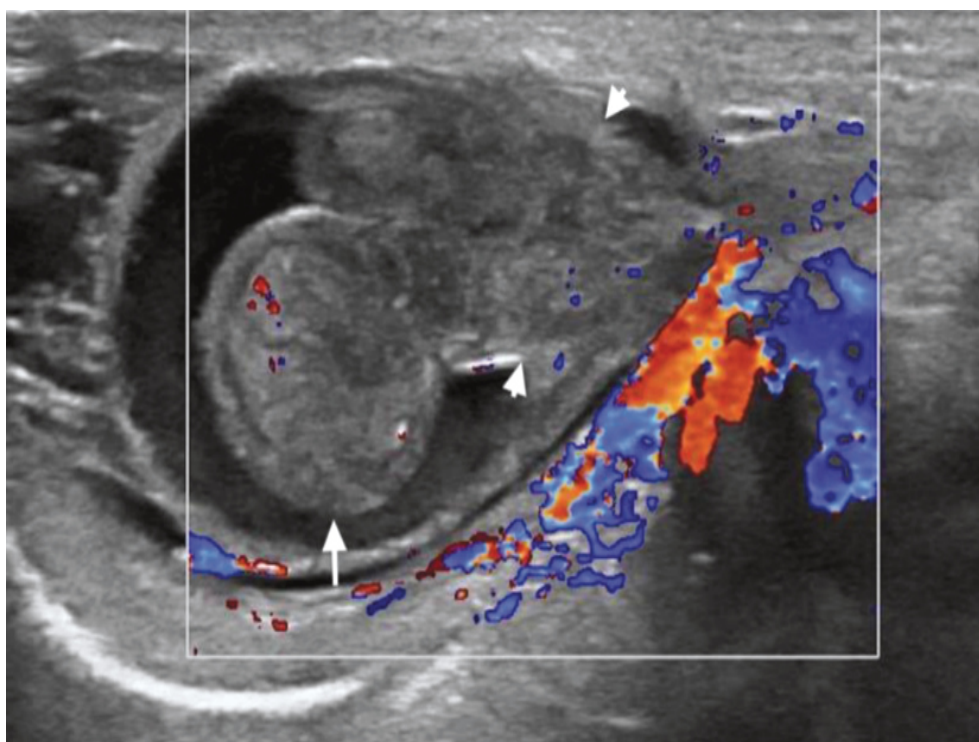


Figure 3 Torsion of the testis in a newborn. CDUS demonstrates no blood flow signal in the testis which has heterogeneous echotexture (arrow). Distal part of the spermatic cord attached to the testis (arrowheads) is enlarged and appears as a hypoechoic tubular structure with no vascularity.

Conclusions

Careful physical and radiological examinations are very crucial for the diagnosis in the neonatal male infants with scrotal discolouration. Especially in the presence of hydrocele on the contralateral side, TT should be considered. Patients diagnosed with TT should be operated as soon as possible, considering the general condition of the patient and the recoverability of the testis. We believe that the fixation procedure should be performed in the same session due to the further high torsion rates in the contralateral testis.

Authors Contributions

TT, MS and MRO designed the study; TT, UB and EG collected and analysed data; TT, MS, UB, and EG wrote the manuscript; TT, MS, UB, EG, MRO and AK gave technical support and conceptual advice. All authors read and approved the final manuscript.

Declaration of Conflicting Interest

The authors declare that they have no any conflict of interest.

Funding

The authors received no financial support for the research, authorship, and or publication of this article.

Ethical Approval

The local ethics committee approved this study (Decision No: 06, Date: 13.06.2019).

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