Neonatal Testicular Torsion: Is it Time for Consensus?

Ranjit I. Kylat

Department of Pediatrics,
College of Medicine,
University of Arizona, AZ,
USA

ABSTRACT

Testicular torsion (TT) occurring in the newborn period has been called neonatal TT (NTT) or perinatal TT. Most of these are asymptomatic, occur prenatally, and are difficult to diagnose. Almost all NTT are extravaginal. There is a lack of consensus in the approach to management of this rare condition. An emergent surgical exploration could salvage, the asymptomatic bilateral TT and prevent asynchronous torsion. After a thorough review of the evidence, the conclusion is that NTT should be managed like TT at any other age group, except that nonoperative maneuvers may not be successful.

KEYWORDS: *Intravaginal testicular torsion, neonatal testicular torsion, orchiopexy, perinatal testicular torsion*

INTRODUCTION

Testicular torsion (TT) in the neonatal period is rare and it occurs in 6.1 per 100,000 live births.^[1] TT leads to testicular ischemia and later to atrophy and if bilateral can lead to infertility and endocrinological dysfunction.^[2] Neonatal TT (NTT) is usually extravaginal, 90% of these occur in neonates and related to lack of fixation and abnormal mobility of the testicle.^[3] NTT occurs during the testicular descent into the scrotum, and many occur in utero.^[3] After the neonatal period, torsion occurs inside the tunica vaginalis - intravaginal torsion, the form of torsion seen in 90% of infants, children and those with undescended testes.^[3] We are presenting an unusual occurrence of intravaginal NTT in the 1st day of life, and there are only three reports of this type in this age group.^[4]

INSTRUCTIVE CASE

A male infant was born at gestational age of 38 weeks to a 19-year-old primigravida. The mother had regular prenatal care, and her pregnancy course was unremarkable. The infant born through vaginal delivery weighed 3955 g (>98th percentile), and head circumference was 36 cm (>90th percentile). The patient had normal vital signs. Examination revealed a left testicular mass at birth which was hard, elevated, slightly enlarged and not trans-illuminant. Abdominal masses were not detected, and the right testicle was descended and normal. He had normal a complete blood count and

electrolytes. Scrotal and testicular ultrasound showed thickening surrounding the left testicle possibly due to thickening of the tunica vaginalis with absent blood flow on power Doppler imaging consistent with a TT. The right testicle measured 1.4 cm × 0.9 cm × 1.0 cm with uniform echotexture without focal abnormality or significant asymmetry. Arterial and venous blood flow was demonstrated by spectral Doppler in the right testicle, along with a moderate right hydrocele. At 18 h of age, when operated on, an unusual intravaginal torsion, with necrotic left testis which had twisted on its blood supply was seen. The patient underwent left scrotal orchiectomy and right scrotal orchiopexy. Pathology revealed infarction and fibrosis. A follow-up visit, 6 months later, did not show any evidence of testicular atrophy on the right.

DISCUSSION

A scrotal swelling in the newborn period is easily missed and could be due to birth trauma, breech delivery, epididymo-orchitis, appendicitis, adrenal hemorrhage, idiopathic scrotal hemorrhage, hydrocele, and TT.^[3] NTT is also called perinatal TT, with most testes undergoing torsion prenatally, and in about 20% it occurs after

Address for correspondence: Dr. Ranjit I. Kylat,

Department of Pediatrics, Division of Neonatal-Perinatal Medicine and Developmental Biology, College of Medicine, University of Arizona, PO Box 245 073, 1501 N Campbell Avenue, Tucson, AZ 85724, USA.

E-mail: rkylat@gmail.com

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birth.^[1,3] NTT is generally asymptomatic, without local tenderness and is difficult to diagnose. Examination findings of NTT generally show a nontranslucent unilateral scrotal swelling with a firm, discolored testicle. Tenderness and absence of cremasteric reflex are unreliable signs in this age group. In some patients, there could be asymptomatic bilateral involvement, at the time of diagnosis (synchronous or metachronous) or later in life (asynchronous). Clinical diagnosis is generally sufficient, but ultrasound using high-frequency linear transducer and color Doppler can be used to exclude a rare differential diagnosis and for the viability of the contralateral testicle, for synchronous bilateral involvement.^[5-7]

There is considerable controversy and variability in the management. The major differences in opinion are related to timing and value of emergent surgery, risks of anesthesia, inguinal or scrotal approach for surgery, occurrence of bilateral synchronous or asynchronous torsion and method of fixation of the contralateral testicle. The grounds for confusion are many, with some contradictory literature, and we review the evidence to date.

Testicular salvage after surgery is low in infancy and was documented in 6% of neonates and in infants up to 17%.^[3] Even when emergent surgery was done within 3 h after diagnosis in a case of bilateral NTT, it could not prevent bilateral testicular atrophy.^[8] In a further review of bilateral torsion, when scrotal exploration was done, on the same day as diagnosis, the authors found that all infants had bilateral testicular atrophy.^[9] They tried to differentiate between urgent surgery done within few hours and early surgery done within days and concluded that the literature does not offer a consensus.^[9] Djahangirian *et al.*, argues that there seems to be no advantage to early intervention and that there is no need for orchiectomy, as torsion leads to ipsilateral testicular atrophy and contralateral orchiopexy should be deferred until the risks of anesthesia and surgery are improved.^[10] Kaye devised a management algorithm and opined that if the findings of unilateral torsion are present at birth, it is likely prenatal in origin and the chance of salvaging the testis is low. He recommended ipsilateral orchiectomy and contralateral orchiopexy after a month of age, through an inguinal approach.^[11]

Lopez and Beasley suggested that “even though the risk of bilateral asynchronous torsion is small, the consequences of bilateral anorchia are devastating and given the relative safety of prophylactic orchiopexy, in an increasingly litigious society, performing contralateral fixation earlier (at 3 months) rather than later was preferred, due to the small but definite risk

of asynchronous torsion”.^[12] Callewaert suggested that immediate surgery is mandatory in suspected bilateral torsions and in cases of possible unilateral torsions that there is no place for a more fatalistic “wait-and-see” approach.^[13] They even suggested that, if a necrotic testicle is found, it should not be removed during surgery, as some endocrine function may be retained.^[13] In another case report of bilateral NTT, the authors suggested that “there was a growing trend of emergency scrotal exploration with contralateral fixation of the testis, as delaying surgery in the postnatal period for prenatally occurring unilateral torsion risks loss of one or both testes, due to a significant risk of bilateral torsion”.^[14] In a retrospective series in Denmark, it was noted that in 85% the testis was nonviable at the time of surgery and in only 10% was it salvageable, even though their conclusion was that “doctor’s delay was common for this rare disease”.^[15] In another literature review, it was suggested that prevention of contralateral TT is paramount and the evidence called for immediate surgical intervention.^[16] In a retrospective review of all case series of NTT, the overall salvage rate was <10%, but when the operation was specified as an emergency, salvage rate seen was 22% with the authors concluding that early surgery with of the contralateral fixation would be ideal.^[17] Roth *et al.* reported that after they adopted a strategy of emergent surgical exploration in cases of prenatal torsion, anorchia was minimized in patients with bilateral asynchronous torsion.^[18]

In a survey of Canadian pediatric urologists, Guerra found that only around 67% would operate for torsion in the neonatal period.^[19] Further, in cases of atrophic testis, none of the respondents opted for immediate surgery, with <40% suggesting, a delayed contralateral orchiopexy. Among the Canadian urologists, a scrotal incision was preferred by only around 50%.^[19] As a commentary to this survey, Salle expressed surprise, that many did not feel that contralateral exploration or fixation was needed and that younger surgeons would operate immediately.^[20] In Synder versus Diamond, opposing expert views of observation and emergent surgery were quite eloquently argued, with the merits of each position carefully analyzed.^[21] Further, in an editorial comment, it was suggested that, if there are no acute changes in an asymptomatic neonate, a recommendation for surgical exploration is not warranted.^[22]

A survey of surgeons and urologists working in the British Isles found that, even though 75% explore the scrotum, only a few surgeons undertake emergent exploration.^[5] Twenty-two percent of surgeons reported that they do not perform contralateral orchiopexy, with concerns of damaging a healthy testicle. Amongst those

who performed it, 50% used nonabsorbable suture fixation and 31% by creating a sutureless extradartos pouch.^[5] A similar survey amongst pediatric urologists in the USA found that in a neonate with prenatal torsion, only 10% would not explore or perform a contralateral orchiopexy. In a neonate with postnatal torsion, 98% would operate urgently, and 96% would perform a contralateral orchiopexy.^[23] The surgical approach preferred was via a scrotal incision.^[23] The wide disparity in these three surveys sheds some light to the ongoing controversies in the management.^[5,18,23] In another expert commentary, Friedman suggest that the risks of general anesthesia during the neonatal period should be considered against the potential devastation of anorchia, should contralateral torsion occur.^[24]

Inguinal exploration to examine for a patent processus vaginalis risks spermatic cord, testicular or vasal injury and the potential for a later development of hydrocele or hernia. Hence most prefer a scrotal approach.^[18,23,25] The testicular workup for ischemia and suspected torsion score, which uses urological history and physical examination to assess risk of torsion, has been validated for older children but is not useful in NTT.^[26] There is no literature in the newborn of successful nonoperative maneuvers being performed.

Reports of intravaginal TT are extremely rare in the neonatal period and early infancy.^[4,27,28] The postulated reasons for this type of TT, are related to excessive laxity of gubernaculum testis and a high attachment of the tunica vaginalis to the spermatic cord.^[4] This could also be related to an anatomical defect of the tunica vaginalis, which can be bilateral. Hence the fixation of the contralateral testis is important to prevent any future torsion.^[4]

The patient described was asymptomatic, and an early physical examination revealed the subtle differences in size and color of the scrotum and testes. The timely intervention did not salvage the affected testicle but likely prevented an asynchronous torsion on the unaffected side. This rare condition reinforces the need for careful physical examination of every newborn infant soon after delivery and for emergent evaluation and surgery for suspected NTT. A further reason for that is that synchronous bilateral TT could be missed on physical examination and only identified on surgical exploration of unilateral NTT. Emergent surgery may result in salvage of the contralateral torted testis.^[18,23,24] Our plea would be that the obviously atrophic testicle is difficult to detect in the immediate newborn period. Neonatal, perinatal and prenatal TT are terms being used, but there is no reason, that emergent treatment, as offered to older infants and children should be

withheld in this population. Currently, neonatal anesthesia is available in all accredited hospitals and the risks of anesthesia are extremely low in neonates with the availability of pediatric anesthesiologists. The consideration of litigation and parental anxiety are all factors some would take in the overall management plan of a patient. However, the paramount treatment plan would be evidence-based care, which now points toward immediate surgery through a scrotal approach with a view to salvaging the affected side, if possible and contralateral fixation. Non-operative maneuvers to reduce NTT are never successful. This is the best way to prevent later anorchia, infertility, and hormonal issues. The questions which are still left to be answered are the method of fixation without sutures or through a dartos pouch.

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Conflicts of interest

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