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Case Report

Neonatal Testicular Torsion Associated with Neonatal Stress: A Case Report

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Abstract

Introduction: Neonatal testicular torsion (NTT) is a rare but critical condition requiring prompt diagnosis and intervention to prevent testicular loss. Though rare, it accounts for greater than ten percent of testicular torsion cases in the pediatric population. **Case Presentation:** A 4-day old, ex-36-week gestational age, male infant was admitted to the neonatal intensive care unit (NICU) for hyperbilirubinemia and choking episodes at home. He was the product of a difficult vaginal delivery associated with clavicular fracture. On day two of admission, a firm left testicle and a right hydrocele were noted. There were no signs of distress, tenderness to palpation, nor scrotal color change during the exam. An ultrasound revealed diminished blood flow to the left testicle with morphology suggestive of NTT and a large right hydrocele. The infant was taken to the operating room where left testicular torsion was confirmed; a necrotic left testicle was unsalvageable. Pathology revealed hemorrhagic necrosis without malignancy. **Conclusion:** Testicular torsion in the newborn period is rare and often difficult to diagnose as there are varying clinical presentations and the infant does not display signs of distress. It has been postulated that fetal stress during pregnancy or delivery is a predisposing factor for NTT; however, little research has been done on whether postpartum stress can contribute to NTT. This case highlights a neonate with testicular torsion discovered following both a stressful delivery and choking event at home. It underscores the importance of considering NTT in neonates with abnormal testicular exam findings, particularly those with stressful deliveries and/or clinical histories.

Keywords: Neonate; Testicular Torsion; Orchiectomy; Stress; Case Report

Introduction

An abnormal scrotal exam in a newborn male can have many causes including genetically abnormal ambiguous genitalia, hydrocele, inguinal hernia, undescended testicle, absent testicle, or testicular torsion. Of the aforementioned diagnoses, neonatal testicular torsion (NTT) is an urgent surgical diagnosis and requires timely management. NTT is a rare event; however, failure to promptly diagnose and treat this event can lead to vascular compromise, ischemia, and testicular loss.[1] Defined as occurring in the first 30 days of life, NTT can have varying presentations depending on the timing of the torsion itself, either early prenatal, late prenatal, or postnatal torsion [1]. The clinical exam findings and therefore likely timing of the torsion are important in determining management as optimal management remains controversial as to whether or not the affected testicle can be salvaged [1,2]. It has been postulated that fetal stress during pregnancy and or/delivery can be associated with NTT and it is known that vaginal delivery, gestational diabetes, and higher birth weight have all been linked to a higher risk of NTT [3].

This case report discusses an infant who was admitted to the neonatal intensive care unit (NICU) for physiologic jaundice and choking episodes who was found to have testicular torsion leading to emergent orchiectomy. This case is noteworthy in that

this infant had known risk factors for NTT including large-for-gestational-age birthweight as well as stressful vaginal delivery, but also a stressful post-delivery course of "choking" episodes, desaturation events, and a failed congenital heart disease screen begging the question if these "choking" episodes contributed to the NTT or if, in fact, were a result of the NTT itself.

Case Report

A 36-week preterm newborn presented to the local emergency department on day of life 4 for an episode of coughing and choking witnessed at home. Pregnancy was complicated by pregnancy induced hypertension. Delivery was notable for a "difficult vaginal" delivery with shoulder dystocia leading to clavicular fracture. His APGARS were 5 and 9 at 1 minute and 5 minutes of life, respectively. His birthweight was 10 pounds, 1 ounce which is considered large-for-gestational age. His first genital exam following delivery was reported as "normal with bilateral hydrocele," but was without further comment. The infant's initial nursery course consisted of a two-day hospital stay significant for a failed congenital heart disease screen. He had a plasti-bell circumcision prior to his discharge from the newborn nursery. There were no other procedures nor concerns during his newborn stay. During his evaluation in the emergency department, his total bilirubin level was 17.2. His exam was significant for jaundice only; vitals were normal.

There was not a scrotal exam documented. Due to his physiologic jaundice and questionable choking episodes, he was transferred to a level 4 NICU via ground ambulance for further evaluation and treatment. During transfer, he was noted to have some desaturation events, but he did not require intervention. On admission to the NICU, his physical exam was remarkable for jaundice and a noted "increased size of bilateral testicles, right > left, without color change or tenderness. Right side transilluminates." This was thought to be a likely hydrocele. He was started on phototherapy and continuous cardiopulmonary monitoring. On the day following admission, physical exam again revealed an abnormal genitourinary exam of "left palpable testicle questionably large with possible surrounding hydrocele though does not transilluminate and feels somewhat larger than normal, no obvious hernia palpable, does not seem to be tender; right hydrocele noted, transilluminates well." A scrotal ultrasound was ordered and revealed a "large right hydrocele; relatively diminished blood flow to the left testicle when compared to the right with abnormal morphology suggestive of possible perinatal testicular torsion; and left complex hydrocele" (Figure 1).

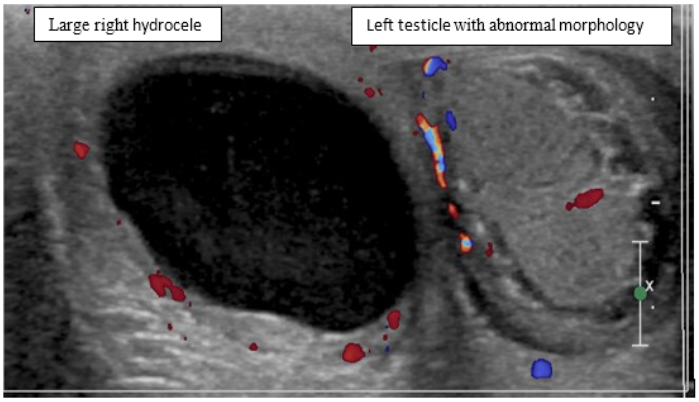

Figure 1: Ultrasound image of large right hydrocele, likely reactive, and left testicle with abnormal morphology and associated hydrocele. Note some preserved blood flow to left testicle.

Urology was consulted, and the infant was immediately taken to the operating room where he was found to have confirmed left testicular torsion with serous sanguinous fluid within the hydrocele sac, left testicle black, and a large right hydrocele (Figure 2). A left orchiectomy and right scrotal orchiopexy were performed. Biopsy showed extensive hemorrhagic necrosis negative for malignancy.

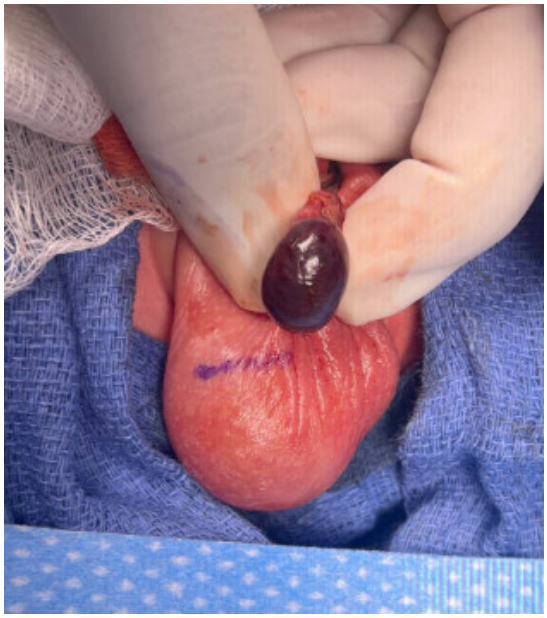

Figure 2: OR image of necrotic left testicle.

The infant tolerated the procedure well. His pain was controlled with morphine and acetaminophen as needed post-operatively. He had an echocardiogram due to his desaturation/choking events that was normal for age. He was discharged three days later without further issues. The infant had follow-up with Urology several months later and genitourinary exam at that time revealed well-healed scrotal incisions and a healthy right testicle without need for further follow-up.

Discussion

Testicular torsion is often thought of as an event that occurs during pubertal years; however, the second peak incidence of testicular torsion occurs in the neonatal period, accounting for 10-12 percent of all cases of pediatric testicular torsion.[1] The mechanism of NTT as well as the associated clinical and surgical findings are quite different from those seen in testicular torsion in other age groups; further, and unfortunately, the outcomes are quite different as well.[3] Most notably, older children and adults with testicular torsion present with an abrupt onset of severe testicular or scrotal pain that is highly suggestive of testicular torsion and leads to urgent evaluation and management[4]; however, NTT is not often associated with obvious infant distress. It is thought that the increased mobility of the neonatal tunica vaginal is within the scrotum during the first month of life as well as an increased cremasteric reflex makes the neonatal testicle more susceptible to torsion along the long axis of the spermatic cord resulting in extra vaginal torsion [1]. This alone, though, is not thought to explain the full breadth of the risk of NTT as it remains a rare event, even among infants born prematurely.

This case is interesting in that this infant was noted to have known risk factors for NTT (namely, larger than average birthweight and a difficult vaginal delivery complicated by shoulder dystocia that ultimately lead to a clavicular fracture), but he also had stressful events after his delivery. First, during his initial newborn course, the infant failed his congenital heart disease screen. Once home, he had "coughing and choking" events described by his parents with associated perioral cyanosis. On his transport to the level IV NICU, the infant had observable desaturation events though he did not require intervention. It must be asked, then, if his coughing and choking episodes at home as well as his unexplained desaturation events contributed to, or, possibly, were symptoms of his NTT. These desaturation events and choking episodes did not persist during his NICU stay and he had none following his diagnosed torsion. No etiology, other than that of prematurity, was ever discovered. It is interesting to theorize that the choking episodes and desaturation events themselves could have been the signs of his distress associated with a twisting testicle. This association, however, has not been described in the literature and cannot retrospectively be proven.

For this infant, once ultrasound evidence of testicular torsion was available, the infant was taken urgently to the OR; however, his testicle was unsalvageable. It is difficult, in this case, to delineate the exact timing of his torsion, largely attributable to the lack of continuity of providers. While his initial genital exam was reported as "normal with bilateral hydroceles," there were different providers who saw him during his initial nursery course, in the ER, on his admission to the NICU, and then again on day 2 of his NICU admission (time of diagnosis). It is known that approximately 70-80 percent of cases of NTT occur prenatally; postnatal torsion is exceedingly rare [5]. By definition, postnatal testicular torsion presents as acute swelling, tenderness, and overlying scrotal changes in a testicle that was previously noted to be normal [1]. This, however, is difficult to determine when provider continuity is non-existent and a normal-to-abnormal physical exam cannot be confirmed. This is quite important, however, as the greatest chance for testicular salvage occurs with postnatal torsion with institutional review studies reporting anywhere from 33-50% [5]. Ultimately, thorough and consistent testicular exams are crucial in capturing cases of neonatal testicular torsion and careful attention should be paid to those infants who are at higher risk. Additionally, providers should consider testicular torsion in males who have unexplained postnatal events, such as oxygen desaturation events in this case, as perhaps these could be symptoms of a torsion testicle.

Conclusion

NTT is a difficult diagnosis, but one that cannot be missed as timely recognition is crucial to optimize outcomes. While NTT is vastly different from testicular torsion in other ages in both pathogenesis and clinical presentation, careful attention must be paid to infants at higher risk for NTT and providers need to maintain heightened clinical vigilance during neonatal testicular exams, particularly in infants who have concerning clinical histories and/or otherwise unexplainable signs or symptoms of distress.

Author Disclosure Statement

There are no financial disclosures to be made or conflicts of interest pertaining to this case report. This case report does not contain a discussion of an unapproved/investigative use of a commercial product/device.

Consent

Informed consent was obtained from the guardian.

Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Conflict of Interest

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