PEDIATRIC SCIENCES JOURNAL
The Official Journal of the Pediatric Department, Faculty of Medicine, Cairo University, Egypt

Case Report

Unilateral Silent Neonatal Testicular Torsion: A Case Report

Ali Jawad[1], Hiba Hamdar[2]*, Najib Hamad[3], Andrea Eid[4], Noama Husseini[5], Ahmad Atieh[6], Abbas Al Bazzal[6], Mohammad Ali Mtairek[1]

[1] Pediatrics Department, Faculty of Medicine, Damascus University, Damascus, Syria
[2] Emergency Department, Notre Dame Maritime Hospital, Byblos, Lebanon
[3] Pediatric Surgery Department, Al-Zahraa Hospital University Medical Centre, Beirut, Lebanon
[4] Pediatrics Department, Faculty of Medicine, Holy Spirit University of Kaslik, Lebanon
[5] Pediatrics Department, Faculty of Medicine, Beirut Arab University, Beirut, Lebanon
[6] Pediatrics Department, Faculty of Medical Sciences, Lebanese University, Beirut, Lebanon

***** Correspondence: hiba.hamdar@live.com

Received: 2/5/2024; Accepted: 3/10/2024; Published online: 25/10/2024

Abstract:

Neonatal testicular torsion is a critical urological emergency that poses a unique diagnostic challenge. It needs a high index of suspicion in a neonate with vague symptoms. It is characterized by the axial rotation of spermatic cord structures, which results in interruption of blood supply to the testicle. The duration of symptoms and degree of twisting have an impact on the viability of the testis, emphasizing the importance of prompt diagnosis and management. We report a 1 day-old neonate who presented by acute scrotum syndrome, with tenderness, hardness, edema, and erythema of the left scrotum. The neonate was afebrile with normal stool passage and without any constitutional symptoms. The neonate was delivered via cesarean section after an uneventful pregnancy. Diagnostic ultrasound revealed an increase in left testicular volume, a markedly heterogeneous structure, and an irregular outline which were considered signs of left testicular torsion. In addition, color Doppler imaging showed the absence of vascularization within the parenchyma, prompting urgent surgery. During surgery, the left testis was found black with no blood supply, so no detorsion was done and the patient underwent an orchidectomy for his left testis. Besides, sympathetic orchidopathia of right testicle was present as edema and redness. Following surgery, a comprehensive approach included a nil per os (NPO) regimen, intravenous fluids, amoxicillin- clavulanic acid, and paracetamol. The neonate was symptom free and discharged on the third day of life. This case highlights that axial torsion can present late as a surgical emergency in the first day of life and emphasizes the need for high index of suspicion in diagnosis and for a prompt multidisciplinary cooperation and intervention to prevent irreversible ischemic damage.

Keywords: neonatal testicular torsion; silent; orchidopexy; diagnosis

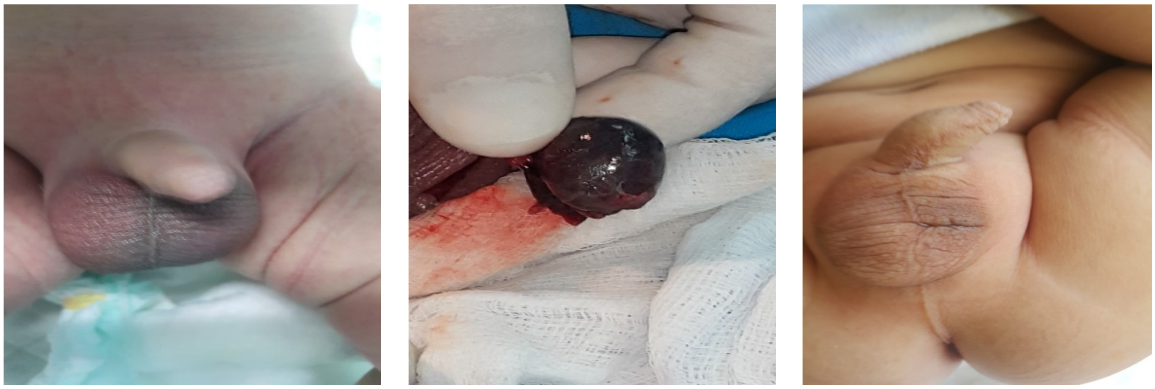
Introduction

Testicular torsion is the rotation of the spermatic cord structures along their axis leading to compression and interruption of blood supply to the testicle on the same side (*1*). Its diagnosis is considered challenging where various other conditions may mimic the presentation of testicular torsion including scrotal trauma, epididymal-orchitis, torsion of the testicular appendage, strangulated hernia, and torsion-detorsion syndrome (*2*, *3*). It is a urological emergency. Prompt identification of testicular torsion and intervention of are crucial for safeguarding the testicle and maintaining future fertility (*4*). Unfortunately, there may be a lack of awareness among medical professionals or parents regarding this urgent urological issue, resulting in delayed diagnosis in most instances. It is widely believed that a testicle twisted for more than 6 hours is beyond the viable time frame from the initial of pain (*5*). Treatment within 6–12 hours yield a 20–50% success rate, and if treated within 12–24 hours, the chance of saving the testicle drops to 0–10% (*1*). Consequently, seeking early consultation with a urological surgeon upon presentation might be crucial, even in the absence of confirming tests (*4*). Neonatal testicular

torsion is a rare event with controversies regarding its etiology, presentation, surgical management and sequelae (*6*). It may be unilateral or bilateral. This report details a rare case involving a male newborn, only 24 hours old, who was admitted to the hospital due to discomfort in his left testicle, presenting with signs of pain, swelling, and inflammation. The assessment confirmed a condition of testicular torsion.

Case Presentation

A 24 hours- old boy was admitted due to evidence of acute scrotum syndrome where tenderness, edema, and erythema were present on the left scrotum. The neonate was delivered via cesarean section after an uneventful pregnancy. Discoloration of the left scrotum was noted at 18 hours of life. The neonate suckled normally and passed meconium within the first 12 hours of life. There was no hospitalization in the NICU prior to the diagnosis. On physical examination, the testes were hard on palpation. Diagnostic ultrasound uncovered compelling findings: the left testicle exhibited increased volume, a markedly heterogeneous structure, and an irregular outline, with Color Doppler imaging revealing a concerning absence of vascularization within the parenchyma. Notably, an artery displayed a curved trajectory around the affected testicle, raising further concerns. The ultimate diagnosis was left testicular torsion, prompting prompt intervention in the form of orchidopexy scheduled on the same day as admission. The surgery was performed 6 hours after the discoloration was first observed, totaling 24 hours from birth. During the operation, the distressing observation of the testes appearing black added a dramatic dimension to the case. Postoperatively, the management strategy encompassed a multifaceted approach. The neonate was placed on a regimen of nil per os (NPO), accompanied by intravenous fluids comprising 20 ml of dextrose 5% and 100 ml of NaCl. Antibiotic coverage was initiated with amoxicillin- clavulanic acid administered at a dosage of 30 mg/kg/dose every 8 hours, and analgesia was managed with paracetamol at 7.5 mg/kg every 6 hours. The meticulous postoperative care extended to the resumption of feeding the day after the operation. Continued vigilance was exercised upon discharge, which occurred on the third day post-admission. The prescribed regimen of amoxicillin- clavulanic acid and paracetamol was advised to be continued for another 3 days, underscoring the importance of ongoing medical surveillance. This comprehensive approach not only addressed the immediate surgical concern but also highlighted the necessity for sustained postoperative management and follow-up to monitor for potential complications or recurrence.



a **b** **c**

Figure 1. Unilateral testicular torsion in a 1 day old neonate.
a) Darkness of left scrotum. b) Black testicle after orchidectomy. c) The left empty scrotum after orchidectomy.

Discussion

Neonatal testicular torsion is defined as torsion occurring prenatally or within the first 30 days of life. The most recognized type in neonates is extravaginal torsion (85%–90%) (*7*), with roughly 70% of cases occurring prenatally (intrauterine) and 30% believed to occur during the first month of life (*8*). Extravaginal torsion, involving the testis, epididymis, and tunica vaginalis twisting on the spermatic cord, is closely tied to the relationship between the tunica vaginalis and the testis (*9*). In normal development, the inferior pole, along with its tunica vaginalis covering, is secured to the scrotal wall by the gubernaculum testis, with these attachments strengthening as the infant grows, typically fully developing by around 3 months of age (*10*).

This development makes extravaginal torsion a condition predominantly affecting early infancy. Fetal and neonatal cases of extravaginal torsion are attributed to the loose attachment between the tunica vaginalis and scrotal wall via the gubernaculum testis, allowing them to twist around the axis of the spermatic cord (*11*).

Prenatal torsion in neonates has been associated with breech presentation and traumatic delivery, potentially leading to increased intrauterine and birth canal pressures, triggering a brisk cremaster response due to loose tunic-scrotal attachments (*6*). Clinical features are often present since birth but may go unnoticed, necessitating suspicion if a firm, non-tender, discolored scrotal or groin mass is observed (*12*). The neonate we report presented late, and the early signs of testicular torsion were lacking. This silent presentation compromised the outcome of the surgical intervention. He did not have systemic symptoms such as fever, lower abdominal discomfort, nausea, and vomiting that were reported to accompany neonatal testicular torsion (*1*). Testicular torsion was difficult to diagnose in our case as he was discharged after birth, as he suckled properly and had no other clinical manifestations, apart from the discoloration that the parents noted by the end of his first day of life. There are multiple diseases that may mimic the symptoms of torsion, hence the usage of Clinical scoring systems can help with decision-making and encourage the prudent use of imaging, without postponing surgery for patients who have a high risk of torsion (*2*). The testicle was lost due to the delayed recognition of torsion, leading to irreversible ischemic damage. It is not clear if this torsion was a post-natal event or was a prenatal condition that presented during the first day of life.

Testicular Workup for Ischemia and Suspected Torsion (TWIST) score was developed based on the clinical presentation; testicular edema (2 points), firm testicles (2 points), absent cremasteric reflex (1 point), nausea/vomiting (1 point), and high-riding testis (1 point) are the five clinical factors that make up this score. A total score of 0–7 is possible. Based on their overall score, patients were categorized as low-, intermediate-, or high-risk (0–2, 3–4, 5–7, respectively) (*13*). TWIST score has 100% positive predictive value (PPV) for the high-risk group and a 100% negative predictive value (NPV) for the low-risk group during the score's validation. They have proposed that patients in high-risk categories can be taken directly for surgical exploration, negating the need for Doppler in low- and high-risk groups (*13*).

Other potential diagnoses for acute scrotum disease in neonates include birth trauma, breech delivery, epididymal-orchitis, scrotal hemorrhage, and hydrocele (*12*). During the physical examination of a suspected testicular torsion, several indicative findings are crucial for diagnosis. A positive Prehn's sign, where elevating the affected testicle does not alleviate pain, is one such key observation. Additionally, Brunzel's sign, characterized by a high-riding and horizontally positioned testis, and scrotal skin retraction, commonly seen in bell clapper deformity cases, are important diagnostic indicators (*14*). Notably, the absence of a cremasteric reflex serves as a highly sensitive marker for testicular torsion, almost confirming its presence.

Several diagnostic methods are utilized, with ultrasonography using Doppler color flow being a commonly preferred option due to its accessibility and non-invasive characteristics (*15*). In our setting, we used this imaging study to confirm the diagnosis. False-positive outcomes can pose a particular challenge in newborns because detecting blood flow in prepubescent testes can be challenging (*14*). Due to the challenges associated with diagnostic uncertainties and prolonged wait times for investigations at certain institutions, many surgeons often rely predominantly on clinical history and examination findings to inform surgical decisions in the majority of cases (*16*).

The approach to managing neonatal testicular torsion is contentious and varies depending on whether the torsion occurred before or after birth (*11*). Prenatal torsion is often considered irreparable due to challenges in detection, the lack of symptoms, and the limited treatment options available (*1*). On the other hand, postnatal torsion is generally regarded as a surgical emergency, with a significant majority of surgeons indicating a preference for prompt surgical intervention. This not only aims to salvage the affected testis but also includes performing orchiopexy on the unaffected contralateral testis (*11*).

Based on the outcomes of physical examination and ultrasound, the patient was diagnosed with left testicular torsion, necessitating urgent intervention in the form of orchiopexy scheduled on the day of admission. While performing the procedure, the alarming sight of the testes appearing black introduced a striking aspect to our case. In any case surgical intervention was mandatory to save the right testicle from sympathetic orchidopathia (*17*) and loss of the other testis.

Testicular torsion necessitates high index of suspicion, timely intervention due to its time-sensitive nature. Typically, there's a narrow four- to eight-hour timeframe before significant ischemic damage sets in, characterized by alterations in testicular histopathology and

detrimental effects on spermatogenesis (*1*). Swift restoration of blood flow to the ischemic testicle is crucial, emphasizing the importance of promptly referring patients to a urologist (*16*).

Unfortunately, in numerous instances, saving the testicle is not feasible due to delayed detection. Parents play a crucial role as their prompt action in seeking medical attention and early diagnosis can potentially lead to the preservation of the testis through immediate exploration (*14*).

Conclusion

This case underscores the critical urgency of accurate diagnosis and management of neonatal testicular torsion. A thorough comprehension of pathogenesis, clinical manifestations, and diagnostic challenges is essential in such a time-critical case. Thus, dealing with neonatal testicular torsion requires a multidimensional approach. This case highlights the importance of early recognition and intervention to prevent irreversible ischemic damage. The lesson learned is clear: healthcare providers should maintain a high index of suspicion for testicular torsion in neonates presenting with scrotal abnormalities, scrotal examination and imaging are paramount for diagnosis and immediate surgical exploration should be considered to salvage the testis. Moreover, the importance of increased parental awareness and a sonic proper medical attention is highlighted, as any delay in intervention is detrimental and causes irreversible ischemic damage. Overall, testicular torsion is a time-dependent challenge that necessitates proper timely, and comprehensive clinical acumen, surgical expertise, and parental awareness.

Author Contributions: All authors searched medical literature, databases, conceptualized, conducted the case review and reviewed the final manuscript. All authors have read and agreed to the published version of the manuscript.

FUNDING

Authors declare there was no extramural funding provided for this study.

CONFLICT OF INTEREST

The authors declare no conflict of interest in connection with the reported study. Authors declare veracity of information.

References

1. Z. Pogorelić, M. Jukić, V. Škrabić, I. Mrklić, V. Fridl Vidas, I. Jurić, D. Furlan, Bilateral Simultaneous Testicular Torsion in a Newborn: Report of a Case. *Acta Medica Hradec Kralove Czech Repub.* **60**, 120–123 (2017).
2. K. R. Qin, L. G. Qu, Diagnosing with a TWIST: Systematic Review and Meta-Analysis of a Testicular Torsion Risk Score. *J. Urol.* **208**, 62–70 (2022).
3. A. Laher, S. Ragavan, P. Mehta, A. Adam, Testicular Torsion in the Emergency Room: A Review of Detection and Management Strategies. *Open Access Emerg. Med.* **Volume 12**, 237–246 (2020).
4. Schick MA, Sternard BT, "Testicular Torsion." in *In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing;* (2024; https://www.ncbi.nlm.nih.gov/books/NBK448199/).
5. A. Saxena, C. Castellani, E. Ruttenstock, M. Höllwarth, Testicular torsion: a 15-year single-centre clinical and histological analysis. *Acta Paediatr.* **101** (2012).
6. M. Riaz-Ul-Haq, D. E. A. Mahdi, E. U. Elhassan, Neonatal testicular torsion; a review article. *Iran. J. Pediatr.* **22**, 281–289 (2012).
7. R. I. Kylat, M. N. Ahmed, Neonatal Testicular Torsion. *Afr. J. Paediatr. Surg.* **19**, 1–4 (2022).
8. J. D. Kaye, S. B. Levitt, S. C. Friedman, I. Franco, J. Gitlin, L. S. Palmer, Neonatal Torsion: A 14-Year Experience and Proposed Algorithm for Management. *J. Urol.* **179**, 2377–2383 (2008).
9. D. Coveney, G. Shaw, J. M. Hutson, M. B. Renfree, The development of the gubernaculum and inguinal closure in the marsupial *Macropus eugenii*. *J. Anat.* **201**, 239–256 (2002).
10. J. M. Hutson, T. Nation, A. Balic, B. R. Southwell, The role of the gubernaculum in the descent and undescent of the testis. *Ther. Adv. Urol.* **1**, 115–121 (2009).
11. A. M. Basta, J. Courtier, A. Phelps, H. L. Copp, J. D. MacKenzie, Scrotal Swelling in the Neonate. *J. Ultrasound Med.* **34**, 495–505 (2015).

12. T. O. Abbas, M. Ali, Bilateral Neonatal Testicular Torsion; Hidden Surgical Nightmare. *Front. Pediatr.* **6**, 318 (2018).
13. C. Manohar, A. Gupta, R. Keshavamurthy, M. Shivalingaiah, B. Sharanbasappa, V. Singh, Evaluation of testicular workup for ischemia and suspected torsion score in patients presenting with acute scrotum. *Urol. Ann.* **10**, 20 (2018).
14. Á. M. Fehér, Z. Bajory, A review of main controversial aspects of acute testicular torsion. *J. Acute Dis.* **5**, 1–8 (2016).
15. F. Arena, P. A. Nicòtina, C. Romeo, G. Zimbaro, S. Arena, B. Zuccarello, G. Romeo, Prenatal testicular torsion: Ultrasonographic features, management and histopathological findings. *Int. J. Urol.* **13**, 135–141 (2006).
16. V. J. Sharp, K. Kieran, A. M. Arlen, Testicular torsion: diagnosis, evaluation, and management. *Am. Fam. Physician* **88**, 835–840 (2013).
17. R. C. Williamson, W. E. Thomas, Sympathetic orchidopathia. *Ann. R. Coll. Surg. Engl.* **66**, 264–266 (1984).