



Bilateral Neonatal Testicular Torsion: A Case Report and Literature Review

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Abstract

Bilateral neonatal testicular torsions (NTT) are exceptional. We report a case of NTT in a five days old male neonate. We had noticed a right blue, and hard testicle, on the clinical examination of a five-day-old male, one day before discharge. He was diagnosed with a right neonatal testicular torsion which was managed by a right orchidectomy and contralateral surgical exploration. Although the left testicle was vascularized on the preoperative Testicular Doppler Ultrasound (TDU), torsion of the left testicle was found peroperatively and treated by detorsion and orchidopexy. Though the left spermatic cord partially regained a red tinge, the testis remained purple, raising doubts about its viability. At two and six-month follow-ups, the left testicle was smooth but small, and a TDU demonstrated that it was poorly vascularized. Hence, in case of torsion, both the testis should be considered.

Introduction

Neonatal testicular torsion (NTT) is defined as testicular torsion occurring before 30 days of age. The incidence of NTT is 6.1 per 100,000 live births,¹ and bilateral cases are exceptional (around 7% of neonatal testis torsions).² Although neonatal torsions are extravaginal,¹ the consequences are as damaging as intravaginal adult torsion. Indeed, neonatal torsion can lead to ischemia and loss of testicular function, such as fertility and testosterone secretion.¹ This rare case concerns a patient presenting with bilateral neonatal testicular torsion.

Case Presentation

Our case was a full-term male newborn who was delivered by an uneventful normal vaginal delivery. The first clinical examination of his genitals in the delivery room was normal. His physical examination on day five of birth highlighted a small, blue, and hard right testicle suggesting NTT. The palpation of the left testicle was unremarkable. A Testicular Doppler Ultrasound (TDU) was performed. (Figure 1)



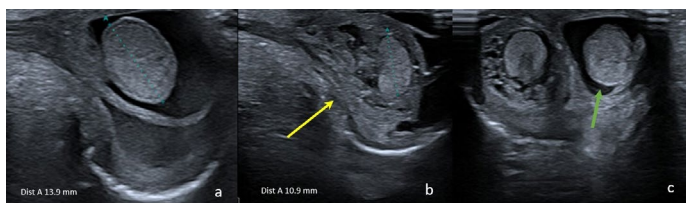


Figure 1: Testicular Doppler Ultrasound. 1a) the left testis is round, homogenous, and well-vascularized. 1b) The right testis is smaller, with a heterogeneous appearance of the peritesticular soft parts (yellow arrow), sphericity loss, and no Doppler flow. 1c) the front view shows the size and echostructure asymmetry of the testicles, and a left hydrocele (green arrow).

The left testicle was 14 mm long, with normal vascularization and a hydrocele. The right testicle was 11 mm long, and doppler showed no vascularization, with a very heterogeneous appearance of the peritesticular soft parts, confirming the diagnosis of unilateral NTT, with hematic effusion. The patient was addressed to the nearest university hospital and hospitalized in the Paediatric Surgery Department. There was no hope to salvage the right testis. However, as the left one was checked twice daily, and seemed normal, and so was the TDU, surgery was planned without any emergency. After three days, he underwent a right inguinal approach and left scrototomy. A right extravaginal torsion was found, with a double-twisted spermatic cord and a purple, stiff, necrotic right testicle. A right orchidectomy was performed, and the testicle was sent for histological study (Figure 2a).

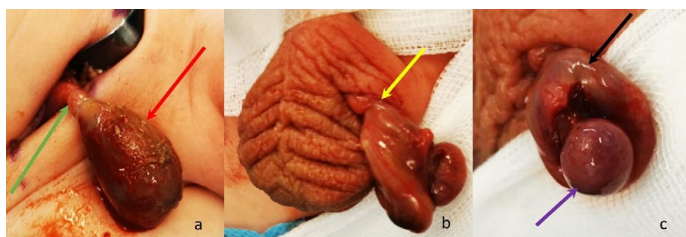


Figure 2: Surgical procedure. a) Right inguinal approach: the spermatic cord is double twisted, with an extravaginal torsion (green arrow). The right testicle is purple, necrotic, and hard (red arrow). b) The left scrototomy (lateral view) highlights an extravaginal testicular torsion (yellow arrow). The spermatic cord is swollen but still vascularized. However, c) on the front view, the left testis is purple (purple arrow), and the spermatic cord is swollen, and weakly vascularized (black arrow). The left scrototomy demonstrated that the spermatic cord was surprisingly one loop-twisted due to a distal extravaginal torsion (Figures 2b and 2c). Most of the spermatic cord was well vascularized, but the left testis and epididymis were purple, though smooth. An orchidopexy was performed using

the Dartos pouch technique. On postoperative day 1 (POD1), the palpation of the left testis was normal. Histological study of the right testis showed a hemorrhagic infarction. At four months old, the clinical examination of the left testis showed a small but smooth testis measuring 7.4 mm. (Figure 3)

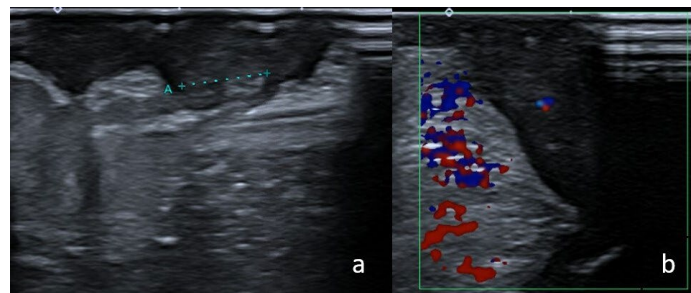


Figure 3: Testicular Doppler Ultrasound at a four-month follow-up. a) The left testis size was 7.4 mm, but homogenous, and the b) Doppler flow was weak.

An endocrine blood check showed a rise in gonadotropin and a low testosterone rate. The patient was then addressed to an endocrinology specialist for further follow-up.

Discussion

This is one of the sporadic cases of bilateral neonatal testicular torsion. NTT is hard to diagnose for many reasons. The incidence rate is low; symptoms can go unnoticed during a routine clinical exam if not acknowledged. Besides, it is not easily detectable on antenatal ultrasound, and the symptoms are quieter (even asymptomatic) than an adult or older boy's testicular torsion. Paediatricians and midwives should consider the rare possibility of NTT if abnormal and atypical male external genitalia is observed during the routine neonate examination, such as swelling, inguinoscrotal bruising, discoloration, or tenderness.¹

As Nandy et al mentioned, bilateral NTT, like our case, with unilateral symptoms, is harder to diagnose.² The TDU's sensitivity in testicular torsion is around 88.9%, and its specificity is 98.8%.³ Although clinical examination and TDU were unremarkable in our case, only surgical exploration established the diagnosis. Hence, we could not know if it was a synchronous or an asynchronous torsion. Because of the one-loop torsion, ischemia might expand slowly, and signs of clinical torsion could not be foreseen. A meta-analysis of 2018 shows that between 8 and 12% of studied patients would have benefited from bilateral exploration and contralateral orchidopexy at the time of diagnosis.⁴ Even if exploration found a non-twisted testicle, asynchronous contralateral testicular torsions tend to be more common than synchronous ones.¹ Consequently, urgent contralateral exploration could prevent

unnoticed testicular torsion or prevent future asynchronous torsion by prophylactic orchidopexy.

Granger J et al presented a case of bilateral testicular torsion, thanks to the systematic contralateral exploration of a first suspected unilateral torsion.⁵ As illustrated by this case and ours, the exploration and fixation of the contralateral testis protect the patient from a future possible torsion and allow the surgeon to track and treat a synchronous torsion. In fact, any torsion may be treated conservatively as much as possible (orchidopexy). Indeed, in animal models, Leydig cells tolerate severe ischemia better than the other testicular cell populations, meaning that testosterone production may still be possible even after a time of anoxia.⁶

Midwives, neonatologists, paediatricians, and paediatric surgeons should treat testicular torsion as a surgical emergency, even if the contralateral testicle seems healthy, because it may not be viable. The risks of recurrence of testicular torsion, testicular atrophy of the lasting testicle, or even decreased sperm count should be explained to the parents.⁷ As reported by Yu CJ et al, follow-up revealed that 56.6% of the boys, who underwent salvage orchidopexy, had testicular atrophy (TA), with median testicular volume loss of the testes around 57.4%.⁸ In our case, the patient showed clinical and radiological signs of TA.

Conclusions

Neonatal testicular torsion is an emergency. Clinical examination suggests the diagnosis and Testicular Doppler Ultrasound can be performed but must not delay the surgical management. However, the risk of testicular atrophy remains high and can challenge the patient's future fertility. Bilateral inguinal exploration is the gold standard procedure, in case of simultaneous or asynchronous testicular torsion. To conclude, we advocate for a mandatory contralateral exploration in an emergency, in any case of neonatal testicular torsion.

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