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CASE REPORT

Perinatal testicular torsion diagnosed in the delivery room: a case report

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Testis,
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Abstract

Introduction: Perinatal testicular torsion (PTT) is a rare condition that most frequently occurs prenatally. Clinical manifestations include abnormalities in testicular consistency, color, or the presence of a groin mass. There are controversies regarding the best surgical approach. The present case report aims to describe a case of perinatal testicular torsion that was approached electively. **Case report:** We report a case of a perinatal testicular torsion diagnosed in the delivery room. The neonate was submitted to elective surgery on the third day of life. Orchiectomy with contralateral orchiopexy was performed. **Conclusion:** In this case report, the neonate was managed by an elective surgical approach, with good evolution. Future studies are necessary to determine the best management for PTT cases.

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INTRODUCTION

Neonatal testicular torsion, also called perinatal testicular torsion (PTT), is a rare condition, with a reported frequency of 6.1/100.000 live births. In most circumstances, it is unilateral and represents around 10% of all testicular torsions in childhood[1]. PTT may occur prenatally, post-partum, or during delivery. Its physiopathology is different from the torsion that occurs in other ages, as it is usually painless and extravaginally (85-90%), which makes diagnosis more difficult[2]. Some risk factors have been described, such as breech presentation, large for gestational-age infants, and difficult delivery[3]. The "*bell clapper*" deformity is a defect in the *gubernaculum testis* that leaves the testicle free to rotate in the *tunica vaginalis*, predisposing to torsion[4].

The rotation of the *testis* around the spermatic cord results in its torsion, which compromises the blood supply and may cause ischemia, which, if prolonged, may result in an irreversible testicular lesion[4].

Clinical manifestations of PTT may include from asymptomatic cases to abnormalities in the testicular color, a firm consistency, or the presence of a groin mass. When the PTT occurs post-partum, it usually presents as local erythema or discoloration, swelling, and tenderness. It is important to emphasize that the cremasteric reflex may be absent at this age, so the absence of the reflex may not relate to PTT[5].

According to the literature, there is great variability in the management of PTT cases. Recent studies have suggested that surgery should be performed immediately, especially to keep contralateral testicular viability[6,7]. However, there are some questions about this emergency approach, particularly regarding the timing of the torsion, and, considering that in the neonate the viability rate of the testis with this approach is only 6%, that surgical procedures have risks and the low chance of being an acute torsion, many specialists suggest that an elective surgery would be the best approach[8].

REPORT

This was a male neonate, birthweight 3,645g (centile 50-90), 40 weeks + 3 days gestational age, 52cm (centile 50-90), adequate for gestational age, born by vaginal delivery. Labor was not prolonged; the 5th minute Apgar was 9 and the presentation was cephalic. No resuscitation was needed. His mother was aged 22 and didn´t present comorbidities. All serological exams during pregnancy were negative.

During the first physical exam, immediately after birth in the delivery room, the neonatologist palpated a painless and hardened scrotal mass, without erythema. An ultrasound was done on the first day of life. The right *testis* showed 1.2 cm^3 and the left 0.4 cm^3 volume. The left *testis* presented a normal shape, normal echogenicity and texture, and normal blood flow by Doppler, with a normal epididymis. The left spermatic cord presented a normal blood flow by Doppler. The right *testis* was diffusely heterogeneous and enlarged, with a very low blood flow by Doppler. The right epididymis and spermatic cord were thickened and heterogeneous, both presenting reduced blood flow. The findings were suggestive of extravaginal testicular torsion.

After the ultrasound, the neonate was evaluated by the pediatric surgeon, who indicated an orchiectomy. Pre-surgery exams were collected, without abnormalities. On the third day of life, the neonate was submitted to surgery, without complications. During surgery, the inspection revealed a hardened right *testis*, wine-colored, with stenosis of the epididymis (Figure 1). The left testis was also explored and a right orchiectomy with contralateral orchiopexy was performed.

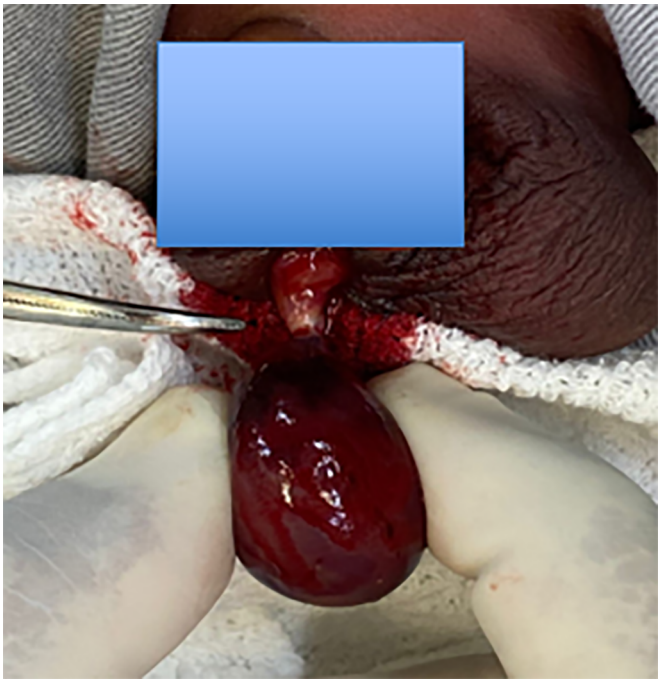


Figure 1. Right *testis* and epididymis stenosis.

The histopathologic analysis revealed a *testis* with 3.0 x 2.2 x 2.0 cm, weighing 6 grams, with a dark and bright external surface suggesting a hemorrhagic infarct. The microscopic exam of the testis, spermatic cord, and epididymis revealed vascular congestion and intense hemorrhage, suggesting a hemorrhagic infarct.

The neonate was discharged on the third post-surgery day in good health conditions and has been followed by specialists. This case report was approved by the Research Ethics Committee (CAAE 57842222.8.0000.5504) and followed the CARE guidelines[9]. Informed consent was obtained.

DISCUSSION

Perinatal testicular torsion is a rare condition, and its surgical treatment should be up to 4-8 hours after the onset of torsion when this time is known. This is called the golden time for surgery. This approach is suggested because salvage rates are higher in these circumstances [5]. When the onset of torsion is unclear, there is variability in surgical management. Some specialists advocate for elective exploration, considering that the rate of testicular salvage in neonates is around 6%, and there are risks associated with anesthesia [10].

In the present case, the neonate presented an abnormal scrotum exam in the delivery room, without signs of acute inflammation, and labor was not prolonged. These factors suggested that testicular torsion has occurred prenatally. In a study with 17 neonates, it was observed that 88% of testicular torsion cases occurred prenatally, suggesting this presentation is more frequent in this age group [10]. Abraham et al. also observed 28 cases of PTT, and this occurred prenatally in 85%[3].

The neonate presented an ultrasound with a right *testis* diffusely heterogeneous, enlarged, and very low blood flow at Doppler. The diagnosis was made on a clinical basis, and confirmed by the ultrasound. It has been reinforced that testicular torsion is a clinical diagnosis and testicular ultrasound with Doppler is the ideal imaging tool to diagnose. A reduced or absent testicular blood flow has a high diagnostic accuracy. The "whirlpool sign" is considered a specific sign of testicular torsion but may be absent[11]. Of note, the presence of heterogeneity with swelling and edema suggests no testicular viability[12].

An elective approach was the choice for the present case. This approach was decided considering the clinical characteristics that suggested that torsion occurred prenatally and was unilateral. In cases where the viability of the testis is probable, or the torsion occurred post-partum, intervention should be done urgently. When the *testis* is viable, detorsion of the affected size and contralateral orchidopexy is the reliable option. In infants, acute testicular torsion is considered an emergency. On the other hand, as the right testis seemed not to be viable at diagnosis in the present case, suggesting an intra-utero torsion, an elective orchiectomy was performed. Perinatal testicular torsion management is controversial, especially regarding the low frequency of testicular salvage. Abraham et al. observed two cases of testicular salvage among 28 PTT cases [3]. The main controversy is regarding the timing of surgical management. DaJusta et al. and Callewaert et al. suggested an emergency approach for all PTT cases, considering the possibility of contralateral testicular torsion and the risk of bilateral anorchia[13,14]. On the other hand, Djahangirian et al in a retrospective study that included 44 neonates suggested there was no advantage to early intervention, and the authors reinforced that the best strategy continues unknown[6].

Our study has limitations. As a case report, we cannot suggest frequency rates. We do not have data about the total number of PTT cases previously observed in the hospital. The strength of this case report relies on the description of the management of this rare condition.

CONCLUSION

In this PTT case report, the neonate was managed by an elective surgical approach, and an orchiectomy and contralateral orchiopexy were performed, with good evolution. Future studies are necessary to determine the best management for PTT cases.

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