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CASE REPORT

Unilateral testicular torsion in a neonate

Torsão testicular unilateral em neonato

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ABSTRACT

A male neonate born at gestational age of 40 weeks was found to have an enlarged and darkened right hemiscrotum after birth. Left testicle was descended and normal. No clinical signs of distress were evident. A color Doppler ultrasound showed an absence of testicular blood flow, consistent with perinatal testicular torsion. The patient underwent a bilateral scrotal exploration through an inguinal incision and a necrotic right testicle was found. A right orchiectomy and left orchiopexy were performed. Perinatal testicular torsion is a rare but severe condition. A high clinical suspicion is required since most of perinatal testicular torsion are intrauterine and can often be asymptomatic, only with localized findings of the affected testis. The management of perinatal testicular torsion is still controversial; however, the most consensual approach is a prompt testicle exploration with orchiectomy of the necrotic testicle and contralateral orchiopexy.

Keywords: Testicular torsion, neonate, testicle, orchiectomy, orchiopexy

RESUMO

Recém-nascido do sexo masculino com idade gestacional de 40 semanas, com edema e escurecimento cutâneo do hemiescroto direito constatados após o nascimento. O testículo esquerdo era palpável na bolsa escrotal e não apresentava alterações. A ecografia escrotal com Doppler revelou ausência de fluxo vascular no testículo direito, achado compatível com torção testicular perinatal. O doente foi submetido a exploração escrotal bilateral através de abordagem por via inguinal, tendo sido confirmada a necrose do testículo direito. Foi realizada orquidectomia direita e orquidopexia esquerda. A torção testicular perinatal corresponde a uma patologia rara, mas com possíveis consequências graves. O seu diagnóstico requer elevada suspeição clínica, uma vez que a maioria dos casos ocorre no período pré-natal, podendo ser assintomáticos após o nascimento e manifestar-se com alterações localizadas ao testículo afetado. A abordagem da torção testicular perinatal é ainda controversa, sendo mais consensual uma exploração escrotal célere com orquidectomia do testículo necrosado e orquidopexia contralateral.

Palavras-chave: torção testicular, recém-nascido, testículo, orquidectomia, orquidopexia

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INTRODUCTION

Testicular torsion (TT) results from the testis rotation around the axis of the spermatic cord attachments preventing blood flow to the testis with consequent ischemia [1]. In extravaginal torsion, the most common type in newborns, a twist of the complete spermatic cord occurs, including the testis, epididymis, and tunica vaginalis [1-3].

TT appears to follow a bimodal distribution, with a first peak in the perinatal period and another later at puberty [4]. Most authors consider perinatal testicular torsion (PTT) to occur prenatally or during the first 30 days of postnatal life, with an estimated incidence of 6.1 per 100 000 births [2,5]. Of all PTTs, about 70% are intrauterine and 30% occur shortly after the delivery [1,2,6].

Due to the potential impact of PTT in the newborn's future fertility, a prompt diagnosis and appropriate management are crucial for preventing irreversible consequences.

CASE REPORT

A male infant was born at gestational age of 40 weeks to a 23-year-old healthy primiparous woman. The pregnancy was uneventful with regular prenatal care and all screening tests were normal. The infant was born through spontaneous vaginal delivery with Apgar scores of 9 at 1 minute and 10 at 5 minutes. His weight was 2820g (15^{th} percentile); length was 48cm (15^{th} percentile) and head circumference was 33cm (3^{rd} – 15^{th} percentile), according to World Health Organization child growth standards [7]. After birth, a firm enlarged non-tender right hemiscrotum was described with slightly darkened skin (**Figure 1**). Left testicle was descended and normal. Clinical signs of distress were absent throughout the hospitalization.

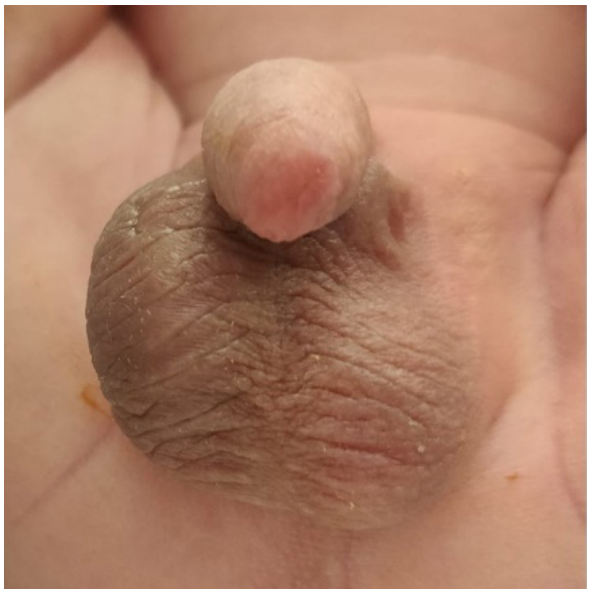

Figure 1 - Enlarged, darkened and elevated right hemiscrotum. Normal appearance of the left hemiscrotum.

A testicular ultrasound was performed and revealed edema, heterogeneous echotexture in half of the right testicle's parenchyma and an irregular outline. There was evidence of thickening of the tunica vaginalis with peripheral hyperechogenic foci and absence of testicular blood flow on color Doppler, consistent with a TT (**Figure 2**). A normal left testicle was described (**Figure 3**).

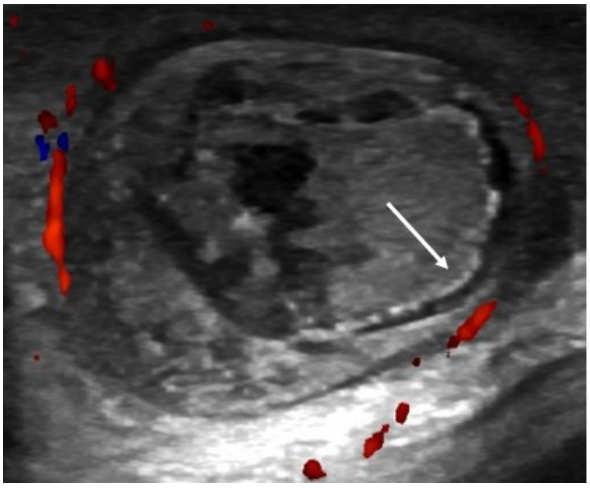

Figure 2 - Sonographic image of the right testicle demonstrating edema and heterogeneous texture of the parenchyma with peripheral hyperechogenic foci (calcifications – arrow) and absence of vascular flow.

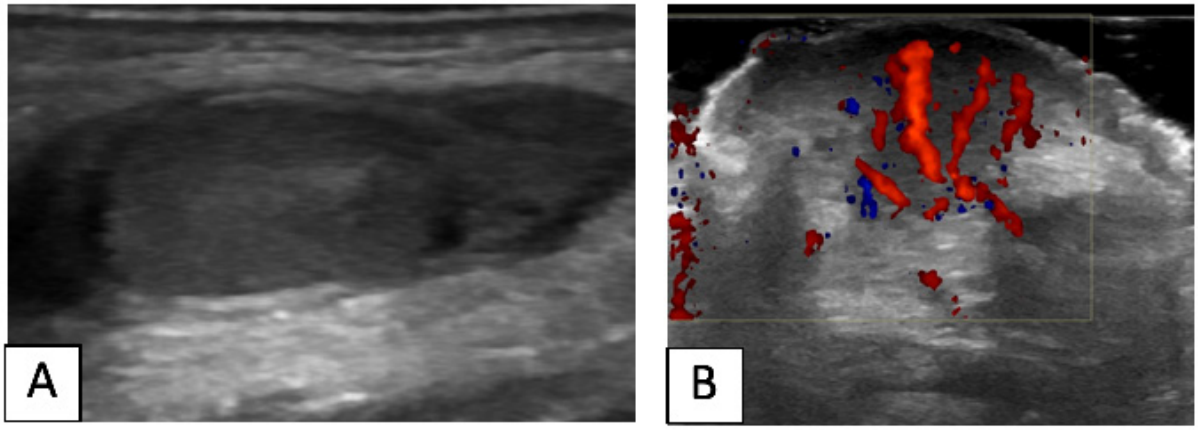

Figure 3 - Sonographic images of the left testicle demonstrating normal size, regular borders and homogeneous texture (A) with normal vascular flow (B).

Given the ultrasound findings, the newborn was transferred to a level III hospital for surgical approach. The patient underwent a bilateral scrotal exploration through an inguinal incision. A necrotic right testicle was observed due to an extravaginal torsion (**Figure 4**). Right orchiectomy and left scrotal orchiopexy were performed. He was discharged the next day, with a follow-up consultation scheduled one month later.

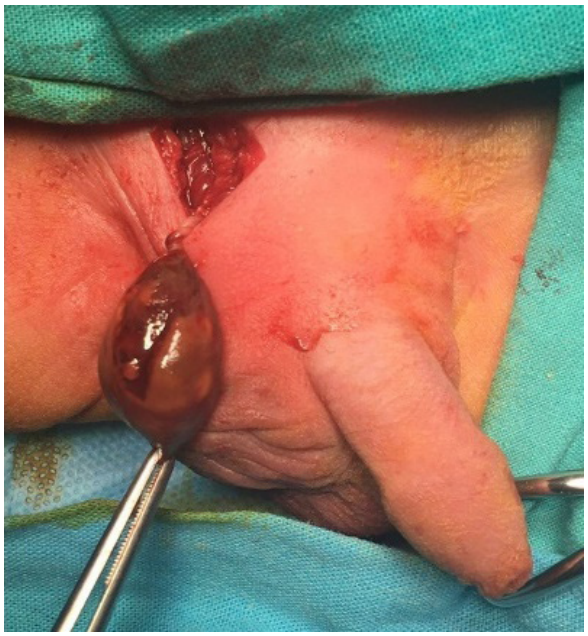

Figure 4 - Necrotic right testicle.

On one-month follow-up consultation, physical examination showed a normal left testicle and an empty right hemiscrotum with normal healing scars. The infant currently has regular follow-up visits with his primary care physician.

DISCUSSION

PTT is a rare urological condition, and it usually occurs unilaterally [8]. TT should be systematically considered as differential diagnosis, as a prompt approach is necessary, especially due to the possible bilateral presentation [1,9]. Bilateral TT have been described in more than 10% of cases and can lead to infertility and endocrinological dysfunction [6,9,10,11].

Although the cause is unknown, some predisposing factors for PTT have been identified, such as breech presentation, prolonged or difficult labor, vaginal delivery and high birth weight [1,3,8,11,12]. The only risk factor in the presented case was a vaginal delivery.

According to the torsion timing, PTT may have different clinical presentations [3]. If torsion occurs after birth, it usually presents as discomfort and the testis will be painful on palpation with classic inflammatory signs, such as swelling and erythema or discoloration [1,6]. The absence of the cremasteric reflex is not a reliable tool for TT diagnosis in infants since the reflex is absent in more than half of healthy children at birth [1,6,9]. Intrauterine torsion presents with localized findings such as a large, firm, discolored and non-tender testis, with no systemic signs

or symptoms [4]. Therefore, clinical diagnosis of PTT is difficult as it can often be asymptomatic [1,9,10]. This case of TT probably had a prenatal origin, which was then confirmed with the testicular ultrasound. B-mode and Doppler ultrasound is used to evaluate testicular morphology and perfusion which can correlate to the chronology of torsion. Generally, heterogeneous testis due to hypoxic-ischemic damage and the presence of calcifications in the transitional zone between the testis and the tunica albuginea are indicative that the torsion may have happened some weeks before birth [6]. Despite the edema of the newborn's testis, it is likely a prenatal TT due to the imaging findings, although the time of occurrence cannot be precisely determined. Ultrasound is also very useful for the assessment of the contralateral testicle viability, for the possible synchronous bilateral involvement, which can also be asymptomatic [9].

Probably in line with its rare occurrence, there is still considerable controversy in the management of PTT [3,9,10].

Some authors suggest that babies diagnosed with prenatal and unilateral TT, due to its low chance of salvaging, should be submitted to ipsilateral orchiectomy and contralateral orchiopexy after one month of age, through an inguinal approach. This would reduce the surgical and anesthetic risks of the neonatal period [3,9,13]. However, the most consensual approach seems to be immediate surgery of the affected side to confirm the diagnosis and try testicle salvaging if possible [3,9,10]. Contralateral exploration is also recommended in order to exclude synchronous torsion and to perform orchiopexy of the unaffected testis because of the risk of asynchronous contralateral TT [1-3,9,12]. There is no place for non-surgical maneuvers to reduce PTT [1,4,9].

It has been suggested that an affected testis should not be removed even if ischemia persists after detorsion because of the postulated higher tolerance to ischemia by Leydig cells [2,10,12]. Some authors argue that some endocrine function may be preserved, maintaining the capacity of endogenous testosterone production, despite the irreversible loss of spermatogenic function [10,12]. This would be of the utmost importance in cases of bilateral PTT. Nonetheless, leaving the necrotic testis in place could be a potential source of infection or carry the risk of tumor formation in the future [2,10,12]. However this issue warrants further investigation [12].

In this case, a bilateral inguinal approach was chosen for the testicular exploration. Nonetheless, the preferred surgical approach for TT is generally via a scrotal incision [1,9,14,15]. It prevents the potential risk of spermatic cord and vasal injury and allows easy access to both testes [9,15,16]. It also avoids the later development of hydrocele or hernia and it is performed in an emergency setting of postnatal TT [1,14].

Notes

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Conflicts of interest disclosure

The authors declare no competing interests relevant to the content of this study.

Authors' contributions.

All the authors declare to have made substantial contributions to the conception, or design, or acquisition, or analysis, or interpretation of data; and drafting the work or revising it critically for important intellectual content; and to approve the version to be published.

Availability of data and responsibility for the results

All the authors declare to have had full access to the available data and they assume full responsibility for the integrity of these results.

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