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CASE REPORT

Perinatal Testicular Torsion - Is it a Rare Occurrence?

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Abstract

Clinical diagnosis of perinatal testicular torsion can be a challenging task as most of the cases are asymptomatic, with no tenderness but may have slight scrotal induration and discoloration. Perinatal testicular torsion has been subdivided into prenatal and postnatal torsion (event occurring from birth to 1 month of life).

Although Doppler ultrasound is frequently used as an adjunct to facilitate the diagnosis of borderline cases, it has its own limitations. Clinical features, treatment options, and fertility outcome depend on when the torsion occurs - prenatally versus postnatally – and when it is discovered.

In this report, we present three cases of unilateral prenatal testicular torsion diagnosed during the routine physical examination prior to hospital discharge and advised to follow up for delayed orchiopexy of the contralateral testicle.

Keywords: Neonatal testicular torsion; Neonatal torsion; Perinatal torsion; Testis; Testicular Torsion;

Introduction

Clinical diagnosis of prenatal testicular torsion at birth is a challenge for all pediatricians and neonatologists as it is not routinely diagnosed by the antenatal ultra-sonogram.[1] Testicular torsion is divided into extravaginal and intravaginal torsion.[2,3] Newborns often present with extravaginal torsion, where the rotation compromises the blood supply. Eventually, it leads to irreversible damage within hours, as the entire testicle including the tunica vaginalis is involved as it is not yet fixed to the scrotal wall. Intravaginal torsion is seen in older children and adults and involves only the testis, which rotates within the tunica vaginalis.[4,5]

According to the consensus-based recommendations of the European Society of Pediatric Radiology Urordiology Task Force and the European Society of Urogenital Radiology Pediatric Working Group, it has been estimated as 6 in 100,000 births, although the exact perinatal incidence rate is unknown.

Seventy percent (70%) of perinatal torsions happen at delivery and only 30% develop postnatally in the first month of life. The overall salvage rate is approximately 9%, and it reaches zero if the torsion was prenatal.[6]

The clinical findings differ depending on whether the torsion has occurred in utero (prenatal) or after birth (postnatal). Postnatal testicular torsion presents with pain, redness, and swelling of the affected testis. This requires acute surgical intervention to restore the blood flow and to prevent the ischemic necrosis, which might occur during salvage of the testis in up to 50% of the cases.[3,5,7] On the other hand, prenatal testicular torsion presents with a hard, discolored, non-tender mass that has already atrophied, with minimal to no discomfort. It is of utmost importance to quickly distinguish between prenatal and postnatal torsion, which merits emergency scrotal exploration to preserve the testis, although the treatment of prenatal torsion is less urgent. A thorough clinical examination and ultrasonography with Doppler to detect the lack of blood flow are the cornerstones in establishing an early diagnosis.[4]

Case presentation

Case 1: A healthy, term male baby delivered with a birth weight of 3375 g by a 21-year-old primigravida mother by spontaneous vaginal delivery at 38+6 weeks of gestation. The baby did not require resuscitation at birth. Mother had an uneventful antenatal period and no past history of maternal medical illnesses.

During routine physical examination at 24 h of life prior to discharge, a hard mass was palpable in the right scrotum with the overlying skin darker in color and non-tender. However, baby was discharged and advised for ultrasound of scrotum with Doppler in the outpatient clinic. Ultrasound with Doppler done on followup visit revealed the presence of the right testicle in the right hemiscrotum and measured 1 x 1.1 x 1.4 cm and was bigger than the left testicle, with the absence of parenchymal vascularity (Figure 1). The left testicle in the left hemiscrotum measured 0.9 x 0.9 x 1.5 cm, was normal in size, and had normal parenchymal vascularity. Pediatric surgeons advised no immediate surgical intervention as the torsion had occurred prenatally, and the testis was already necrotic and non-viable. The need for delayed left orchiopexy was explained to the parents.

Case 2: A healthy, term male baby delivered with a birth weight of 3624 g by a 24-year-old primigravida mother by spontaneous vaginal delivery at 40+3 weeks of gestation. The baby did not require resuscitation at birth. Mother had an uneventful antenatal period. No history of gestational diabetes mellitus or pre-eclampsia. Baby was born through a meconium-stained amniotic fluid and was admitted to NICU for observation due to low cord pH of 7.01. During the NICU stay, the baby had normal sensorium with good cry, tone, and activity. Moro and suck reflexes were present.

During the routine physical examination prior to discharge, a firm, small, non-tender left testis was detected with no edema or discoloration of left scrotum. The right scrotum had minimal hydrocele with normal consistency of the right testis. An urgent Doppler ultrasonography of the scrotum revealed the absence of parenchymal vascularity in the left testicle which measured 1.1 x 0.8 cm, suggestive of left testicular torsion (Figure 2). The right testis was normal in size and position, measured 1.2 x 0.7 cm, and showed parenchymal echo pattern and vascularity with mild, clear hydrocele. Pediatric surgeons advised no immediate surgical intervention as the torsion had occurred prenatally and the baby was asymptomatic. Baby was discharged to follow up in the clinic as outpatient for delayed right orchiopexy.

Case 3: A healthy, term male baby delivered with a birth weight of 3785 g by a 38-year-old G6P5A1 mother by spontaneous vaginal delivery at 40+2 weeks of gestation. Baby did not require resuscitation at birth. Mother had an uneventful antenatal period. No maternal history of risk of early-onset sepsis. No history of gestational diabetes mellitus or pre-eclampsia.

During the routine physical examination, the left scrotum was found to be empty, giving the clinical impression of left undescended testis. On palpation,

a nubbin mass at the root of the left scrotum and minimal hydrocele in the right scrotum with normal consistency of right testis was noted. An urgent Doppler ultrasonography of the scrotum revealed absent left testicle in the scrotal cavity, which had a small echogenic, ovoid structure measuring 0.4 x 0.3 x 0.5 cm in the proximal inguinal canal, suggestive of atrophied left testis (Figure 3). The right testicle was located in the right hemiscrotum and had a normal size, measuring 1.1 x 0.7 x 0.9 cm with normal echogenicity and vascularity. Pediatric surgeons advised no immediate surgical intervention as the torsion had occurred prenatally and the testis had atrophied to a nubbin mass and also the baby was asymptomatic. The baby was discharged to follow up in the clinic as outpatient for delayed right orchiopexy.

Investigation

The images of the Doppler ultrasound of the scrotum for all three cases of neonates with the clinical suspicion of prenatal testicular torsion are shown below.

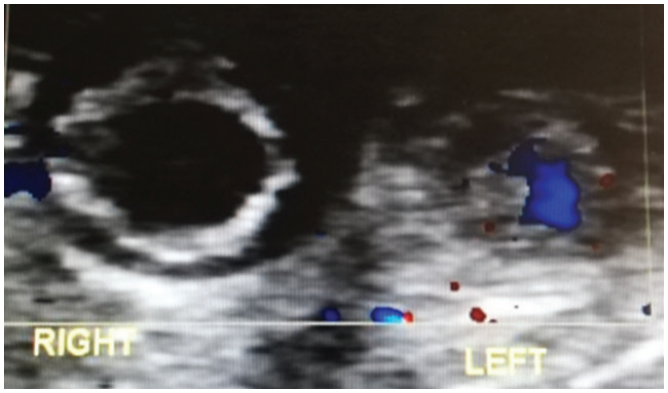

Figure 1: Doppler ultrasound of scrotum showing absence of parenchymal vascularity in the right testis.

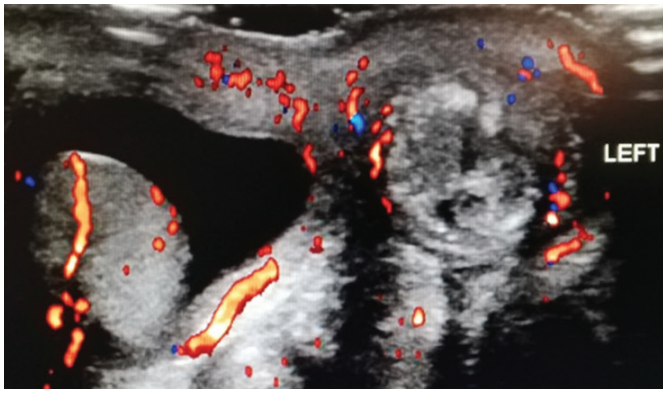

Figure 2: Doppler ultrasound of scrotum showing absence of parenchymal vascularity in the left testis.

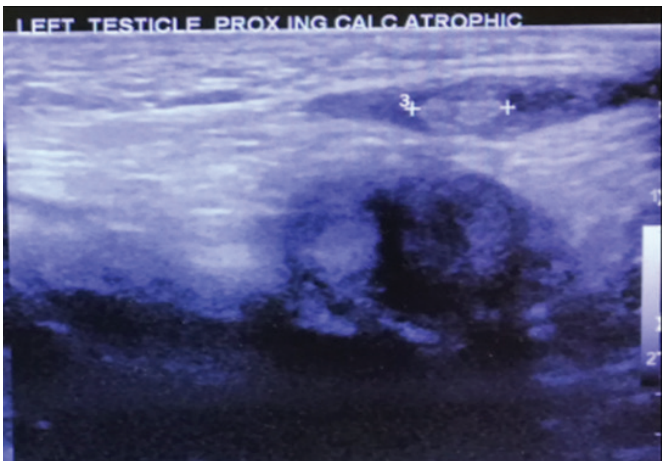

Figure 3: Ultrasound of scrotum showing small nubbin left testis in the proximal inguinal canal.

Treatment

The treatment option recommended for prenatal testicular torsion is delayed orchiopexy of the contralateral testis in order to prevent late asynchronous contralateral testicular torsion.

Outcome and Followup

It has been shown in the literature[6] that the salvage rate of prenatal testicular torsion is almost zero percent. For longstanding intrauterine testicular torsion, surgeons suggest delayed orchiopexy of the contralateral testis and removal of the affected testis (orchiectomy) due to a theoretical risk of tumor formation. The three cases reported here were managed conservatively with less urgency for surgical exploration and advised for follow up in the outpatient clinic for delayed orchiopexy.

Discussion

Prenatal testicular torsion is an unexpected finding for neonatologists and parents, irrespective of the type or mechanism of torsion. It is often diagnosed during the routine postnatal physical examination, as the antenatal ultrasound is not sensitive in detecting this abnormality.

A review of the urological literature demonstrates no consistent pattern with regard to the potential etiologies of perinatal testicular torsion. Possible theories explained include difficult and traumatic labor, breech presentation, large-for-gestational-age babies, an over-reactive cremasteric reflex, pre-eclampsia, gestational diabetes mellitus, and multiparity.[8] However, none of the cases in our study had any of these predisposing factors.

The clinical feature of testicular torsion depends upon when the torsion occurs, either prenatally or postnatally.

In this report, we observed that all the three cases presented at birth had prenatal testicular torsion, but the torsion had occurred at different time periods of pregnancy. A careful clinical examination at birth will help us to differentiate the timing of prenatal torsion, which may not always be detected by the Doppler ultrasound of the scrotum.

If prenatal torsion occurs during the *early part of pregnancy,* far from birth, the newborn will be born with an absent or a nubbin testis. Our case 3 had a similar clinical picture, with an empty left scrotum with a nubbin testis detected on palpation in the proximal inguinal canal and confirmed with Doppler ultrasound.

If torsion occurs *several weeks prior to birth,* the newborn will present at birth with a regular, firm, painless scrotal mass, often in the upper part of the hemiscrotum, which is smaller than the contralateral normal testis. Usually, it is not associated with acute inflammatory signs and does not transmit light. Our case 2 presented with a similar clinical picture.

If torsion occurs *several days prior to birth,* the newborn will present at birth with a firm and painless scrotal mass, bigger than or similar in size to the contralateral normal testis. Our case 1 presented similarly with a hard right scrotal mass which was slightly bigger in size than the contralateral normal testis and without acute inflammatory signs.

If torsion occurs *very near to birth, i.e., several hours prior to birth,* the newborn will be born with acute inflammatory scrotal signs: a painful, enlarged, bluish or reddish hemiscrotum with an enlarged testis; however, it does not transmit light and is with a thickened and painful cord.

If the torsion occurs in the postnatal period within the first month of life, the newborn will be without scrotal signs (occasionally hydrocele), and the acute inflammatory signs will appear later.[9]

Controversy still persists regarding the optimal management of clinically diagnosed perinatal testicular torsion. Management of prenatal testicular torsion has a lack of consensus in terms of surgical timing and need for contralateral fixation.[1]

Cuervo et al[9] managed their patients by considering whether the torsion was long-standing intrauterine, very near delivery, or postnatal. Their study concluded that for long-standing intrauterine torsion, there is no need for urgent intervention and these neonates can be operated electively when the child is in optimal clinical status. This gives enough time to confirm the suspected diagnosis in order to remove the affected testis and to explore the contralateral normal one. According to them, if torsion occurs in the prenatal period very near to birth or in the postnatal period within the first month of life, immediate exploration should be carried out.

Few reports are available on the rate of testicular salvageability in this group of patients secondary to the established infarction.[10]

Is contralateral orchidopexy justified? It is a controversial issue. Study by Kashif et al[11] mentioned 11 cases of neonatal testicular torsion and the contralateral testis was fixed in all cases. Similarly, another study by Mishriki et al[12] was also of the opinion that the contralateral testis should be fixed whatever the cause may be. Their studies concluded that the affected side should be explored promptly to confirm the diagnosis and to fix or remove the affected testicle, and the contralateral scrotum should be explored for orchiopexy because of the risk of asynchronous contralateral testicular torsion.

Conclusion

- Three cases of prenatal testicular torsion had been reported in 6 months, which makes us re-think - Should perinatal testicular torsion still be considered as a rare occurrence?
- A careful clinical examination prior to discharge of all newborns is the key for diagnosis of both prenatal and postnatal testicular torsion, which can be potentially devastating if missed.
- The clue to clinical diagnosis is the consistency of the testicular mass which can be firm-to-hard on palpation with no acute inflammatory signs in case of prenatal testicular torsion.

- More cases should be reported in order to determine the actual number of occurrences of prenatal testicular torsion as this is not an uncommon entity.

Conflict of interest

The authors declare that they have no competing interests.

Acknowledgment

James J prepared the manuscript. Emad Shatla and Minoosh Nasef reviewed the manuscript. Hussain Hamdy did the surgical followup of all the cases. We are indebted to Prof. Imelda Lambert for her support in writing the manuscript.

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