



A Rare Neonatal Testicular Swelling Cause: Neonatal Testicular Torsion

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Abstract

Testicular torsion is a rare medical condition among neonates. Main symptoms are swelling, erythema and pain of the scrotum. It can be both perinatal (diagnosed at birth) and postnatal (diagnosed after birth to 28 days of life). If not treated, atrophy of both involving and/or contra lateral testis can be developed. Still being the main treatment option is surgery, new researches suggest delayed surgery in perinatal torsion because of low rates of salvaging involving testis. We present a perinatal testicular torsion newborn who undergone surgery in third hour of life. Orchiectomy of the effected testis and orchiopexy of the contra lateral testis is performed. To the best of our knowledge, this is the earliest diagnostic time report of a testicular torsion with successful management. Physicians should always alert for testicular torsion in neonatal period, even in delivery room on performing first physical examination of the newborn baby.

Received: Sep 25, 2022

Accepted: Oct 18, 2022

Published Online: Oct 20, 2022

Journal: Annals of Pediatrics

Publisher: MedDocs Publishers LLC

Online edition: <http://meddocsonline.org/>

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Introduction

Perinatal testicular torsion is a rare condition that can be seen in first month of the life. It can be divided into two parts; prenatal which is diagnosed at birth or postnatal which is diagnosed after birth but before 1 month of age. It's extremely rare and its etiology is not well understood. However association between preeclampsia, gestational diabetes, large of gestational age, hydronephrosis and prolonged delivery are suggested [1]. Also in some cases which seen both of the siblings suggests that there might be a genetic inheritance [2].

Testicular torsion is defined as rotation of the testis. It spontaneously rotates around its longitudinal axis, which results twisting of the spermatic cord and lack of blood flow to testis. If

reduction cannot be made immediately permanent dysfunction and infertility can be seen. It can be either unilateral or bilateral. Two types of testicular torsion is defined: Extravaginal torsion involves both testis, epididymis and tunica vaginalis twist of spermatic cord and intravaginal torsion is defined as rotation of the testis without tunica vaginalis [1].

Urgent medical care is previously suggested in both prenatal or postnatal [3]. However new researches question the need of urgent surgery for prenatal torsions [4]. In this case report we present a unilateral prenatal testicular torsion newborn case that defined as torsion of the testis in delivery room after the birth and undergone urgent surgery in first hours of the life.



Cite this article: Bozat AD, Baldemir N, Bahadir GB, Calisici E, Yaprak D, et al. A Rare Neonatal Testicular Swelling Cause: Neonatal Testicular Torsion. *Ann Pediatr.* 2022; 5(2): 1106.

Case report

A newborn baby was delivered in delivery department of our institution from a 44-year-old mother (gravida 1, para 1- in vitro fertilization pregnancy) with cesarean section with birth weight of 3060 gr. His gestational week was 36 wk and 6 days. Mother had polyhydramnios and gestational diabetes with diet regulation during the pregnancy. Upon birth, the newborn baby's 1 and 5th minute APGAR scores were 9/10 respectively. After the few seconds of birth, in delivery room, pediatric resident realized that baby's right scrotum was swollen and larger than the left testis. An erythema on the right scrotum was also present on physical examination (Figure 1). Pediatric surgeons immediately invited to the delivery room and their examinations suggested testicular torsion of the scrotum. Doppler ultrasonography revealed no blood flow to right testis and torsion of the right testis. In his 3rd hour of life, right orchiectomy and left orchiopexy was performed (Figure 2). There were no problems both peri and postoperative period. After the surgery he admitted to our neonatal intensive care unit and had an uneventful followup.



Figure 1: Swelling and erythema of the right scrotum.

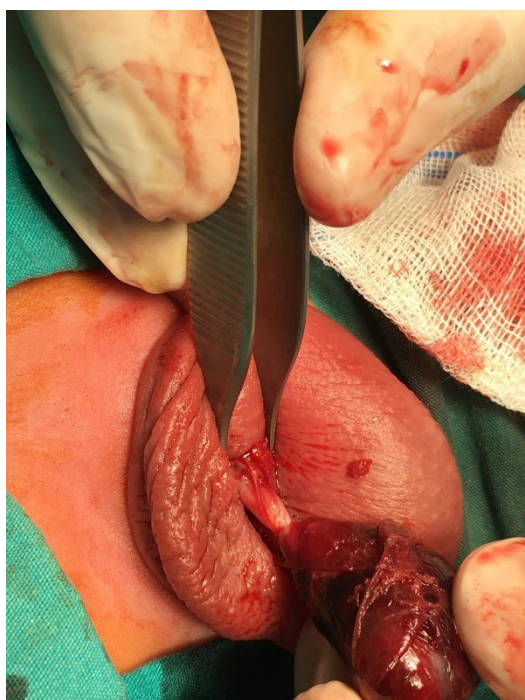


Figure 2: Intraoperative picture of right testis before untwisting.

Discussion

While testicular torsion is a common medical problem in pediatric population, mostly seen in puberty, the incidence of neonatal testicular torsion is a rare medical condition. In neonatal population its estimated incidence is 6.1/100,000 newborn. Neonatal testicular torsion differs from torsion in older children, as it occurs extravaginally, where the tunica vaginalis is not yet fixed to the dartos, and torsion involves the testicle and tunica twisting within the scrotum. Testicular salvage rates are negligible, presumably due to a prolonged ischemic period, secondary to a late diagnosis or torsion prior to delivery [5,6]. In our case a thorough examination in delivery room revealed the early diagnosis of the testicular torsion.

There have been multiple etiologic theories regarding why torsion would occur. It appears that intrauterine stress may be a significant risk manifested by prolonged or difficult labor, high birth weight breech presentation, preeclampsia or vaginal delivery to name a few described in previous studies [4,7]. Also there are some cases that suggests that there might be a genetic pattern [2]. Insulin like peptide-3 (INSL3) and Relaxin family protein receptor 2 (RXFP2) are most suspected genes [8]. But still no correlation has been shown between genes and testicular torsion. It has been postulated that hypermobility of the tunica vaginalis within the scrotal sac when exposed to an extreme cremasteric reflex during deliver or in utero, may induce a torsion. Although, vaginal delivery was found to be associated with neonatal testicular torsion, our case was born with c-section. Preeclampsia of the mother may cause intrauterine stress as a possible contributing factor for testicular torsion in our infant [5,7,9].

Because of rarity of neonatal testicular torsion, the proper management has been difficult to ascertain. Currently, the most common practice is to perform bilateral exploration with orchiopexy of the unaffected testicle to prevent the devastating effects of anorchia, should asynchronous torsion occur [10]. Due to presence of swelling in scrotum upon birth our case was diagnosed as perinatal testicular torsion and urgent right orchiectomy and left orchiopexy was performed. Both surgery timing and surgical strategy is still debatable upon pediatric surgeons. Most of these cases effected testis is not salvageable because of prolonged time of torsion. For this reason, orchiectomy of the effected testis and orchiopexy of the contra lateral testis is still suggested way of treatment. This is because to prevent hormonal activity rather than preventing infertility. Leydig cells are much more resistant to hypoxia there is a chance to restore their hormonal capacity but not fertility, thus while spermatogenesis is diminished a rather normal activity in Leydig cells can be seen after prolonged torsion [6,11].

Timing of operation is another problem in neonatal testicular torsion cases. While in our case a surgery was performed in early three hours in life there are still researches that suggest a delayed operation. Main reason for this is high risks of operative complications and near zero percent salvaging of the effected testis. However due to advances in both neonatal intensive care and pediatric anesthesia, complications are relatively low. In literature there are testicular torsion cases that had been operated as early as 24 weeks without any complications [12]. A study performed by Mayo Clinic revealed a ratio of 0.3 anesthesia related cardiac arrest in children in 10,000 non cardiac surgeries. This ratio was four in 1,014 anesthetics in neonatal population [13]. Also, there are some new information in salvaging rates too. In a meta-analysis by Monteilh et al. in 2019, nine studies

were reviewed and 196 neonatal testicular torsion patients were pooled. In this analysis 156 patients were undergone bilateral exploration. Salvaging of the ipsilateral testis was 7%. Also, they stated that 4% of all preoperative unilateral testicular torsions were bilateral. With this combined result they stated that around 8-12% of the patients would benefit from bilateral exploration [10]. Roth et al reported three cases of ipsilateral prenatal torsion that found to be bilateral at the time of surgery [6,10]. With these information many researchers suggest urgent surgical approach in perinatal torsion, however a guideline was not been established yet. Despite low salvaging rates of the effected testis neonatal testicular torsion should be considered as a medical emergency and urgent surgery should be performed to salvage the effected testis or preventing bilateral torsion and anorchia [4].

True effect of testicular torsion on fertility is not well understood. Around 0.5% of infertility patients had a history of testicular torsion. However, researches evaluating semen in patients with history of testicular torsion showed abnormal results but nor significant results. Also, there were no statistically differences comparing with other causes of monorchia [14]. In a study by Arap et al showed no difference in sperm count and mobility in patients who had testicular torsion for no longer than 24 hours in childhood. Anti-sperm antibody levels were higher but not statistically significant [15]. However, the effect of neonatal testicular in infertility is not fully understood. These results suggest that testicular torsion might have a less contribution to infertility than expected. In our case a pediatric endocrinology follow-up was scheduled to assess the fertility.

Conclusion

In conclusion, neonatal testicular torsion that occurs within the perinatal period is very rare. Any suggestion of viability urgent exploration should be considered at the time of the diagnosis. This treatment modality is not only in the attempt to salvage the affected testicle but to prevent the devastating effects of a bilateral torsion with subsequent anorchia.

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