PEDIATRIC UROLOGY
Case Report

# A case of bilateral perinatal testicular torsion that presented with unilateral torsion; necessity of contralateral testis exploration

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## ABSTRACT

Perinatal testicular torsion is a rare emergency in a neonate that prompts immediate attention. Bilateral testicular torsion is extremely rare. We report a case of bilateral torsion that presented with unilateral scrotal swelling but significant atrophy and dark discoloration of the contralateral testis that was secondary to asynchronous prenatal torsion. There is no consensus about exploration of the contralateral testis when exploring a case with unilateral testicular torsion. Nevertheless, findings in this case report indicate that bilateral exploration is mandatory in each case of perinatal testicular torsion to evaluate the condition of contralateral testis and fix it to prevent development of future torsion that may result in anorchia.

**Keywords:** Neonatal; testicular; torsion.

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## Introduction

Perinatal testicular torsion is defined as testicular torsion that is diagnosed at birth (prenatal testicular torsion) or occurs during the first month of life (neonatal testicular torsion).[1,2] This is a rare entity and its etiology is not well understood, however some associations have been suggested, including preeclampsia, gestational diabetes, twin gestation, large size for gestational age, presence of prenatal hydronephrosis or prolonged delivery.[2,3] In addition, there may be a role for genetic factors, as this entity has been also reported in siblings.[4,5]

It has been suggested that exclusively extravaginal twisting of the spermatic cord which is due to poor adhesion of the tunica vaginalis to the gubernaculum involves in the pathogenesis of the perinatal torsion. As a result, testis and tunica vaginalis twist together as a unit and induces testicular asphyxia.[3,6,7] Despite that, intravaginal testicular torsion is associated with a typical deformity in which testis is not fixed in the tunica vaginalis and can twist freely on its pedicle like a a bell clapper. This type of torsion is called intravaginal torsion which is frequently seen in prepubertal boys. Intravaginal torsion has been reported in an undescended testis of a 30-day newborn boy.[8]

Although previously emergent exploration in all cases with neonatal torsion was recommended, [1,9,10] the advantage of this approach for the newborn baby has been questioned recently.[2] Nevertheless, we report a case with unilateral presentation of testicular swelling and pain with normal appearing contralateral testis that was finally diagnosed as bilateral torsion during exploration. This case supports emergent exploration in these cases to evaluate the condition of contralateral testis and fix it to prevent future torsion that may end in anorchia.

## Case presentation

A boy was referred to our emergency department for urologic consult at postnatal 12$^{th}$ hour. He

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**Table 1. Results of some reports of perinatal testicular torsion**

| Author, date | Number of cases | Presentation | Result of exploration | Decision for symptomatic side | Contralateral testis | Follow up | Recommendation for emergency exploration | Recommendation for contralateral testis exploration |
|---|---|---|---|---|---|---|---|---|
| Van Glabeke et al.[16], 2000 | 18 | Unilateral | Unilateral | Orchidectomy in 17 cases, one case testes preserved | Pexed | | Yes | Yes |
| Olguner et al.[17], 2000 | 1 | Bilateral | Bilateral | Right testis Orchidectomy and left testis detorsion | | | Yes | Yes |
| Arena et al.[18], 2005 | 1 | Bilateral | Bilateral | Two testis detorsion | | | Yes | Yes |
| Al-Salem AH.[19] 2007 | 11 | Unilateral | 10 cases Unilateral and 1 bilateral | In 7 cases orchidectomy and other testes preserved and follow | Pexed | Ischemic Were viable | Yes Preserved testes Excepting one case with atrophy | Yes |
| Ahmed et al.[20], 2008 | 2 | Unilateral | Bilateral* | | Pexed | | Yes | Yes |
| Ralahy et al.[21], 2010 | 1 | Unilateral | Unilateral | Orchidectomy | Pexed | | Yes | Yes |
| Sachwitz et al.[22], 2012 | 1 | Bilateral | Bilateral | Bilateral orchidectomy | Pexed | | Yes | Yes |
| Drlik et al.[13], 2013 | 1 | Unilateral | Bilateral* | Unilateral orchidectomy | Pexed | Necrosis of the | Yes Preserved testis | Yes |
| Granger et al.[14], 2016 | 2 | Unilateral | Bilateral* | Unilateral orchidectomy in two cases | Pexed | No atrophy in | Preserved testis | Yes Yes |
| Abraham et al.[12], 2016 | 28 | Unilateral | 3 bilateral cases* | | | Pexed | | Yes Yes |
| Kawamura et al.[23], 2016 | 1 | Unilateral | Unilateral | Orchidectomy | Pexed | | Yes | |

*Incidental finding during exploration of a case with contralateral testis torsion

weighed 3.600-g, and, the second baby of a 30-year-old woman (gravida 2, para 2) with history of gestational DM and hypothyroidism. Routine prenatal sonogram obtained at the 36th week of gestation was normal, so repeat cesarean section had been planned and performed at the 38th week+2days of gestation.

Pediatrician referred the neonate to us because of erythema and swelling of the left side of the scrotum associated with agitation and persistent crying. In physical examination, the scrotal wall was erythematous at the left side, the left testis was tender and bigger than normal, and we were not able to differentiate the epididymal margin. The right testis was small but normal in physical examination. Sonographic evaluation showed heterogeneous pattern of the left testis and mild hydrocele at the right side. Further, both testes showed no perfusion in color Doppler sonography. The patient underwent surgical exploration immediately. The left testis was edematous and necrotic and a 1080° extravaginal torsion was apparent (Figure 1). The right testis was atrophic and dark colored because of recurrent prenatal intravaginal torsions. The testis had no connection to the scrotal soft tissue that is the typical bell clapper deformity (Figure 2). The left necrotic testis was removed. The right atrophic testis was preserved because of possible endocrine function and was fixed to scrotal wall to prevent torsion. The postoperative course was uneventful. After four months, the right testis was completely normal in physical examination. Written informed consent was obtained from parents of the patient for publication of this case report and relevant images.

Figure 1. The left testis was edematous and necrotic and a 1080° extravaginal torsion was apparent

Figure 2. The right testis was atrophic and dark colored because of recurrent prenatal torsion

## Discussion

In case of perinatal testicular torsion, surgeon should weigh the risk of anesthesia in a newborn against the possibility of preserving testicular function. Previously most specialists validated early surgical intervention,[1,9,10] while recently some experts do not recommend emergent exploration for these patients since in their experience none of the cases had derived any benefit from early exploration.[2] Nevertheless, boys with severe pain and agitation associated with scrotal swelling and erythema and tenderness favouring recent testicular torsion, like our patient, should undergo testicular exploration to relieve pain, to assess testis viability and to fix contralateral testis so as to prevent subsequent anorchia.

Regarding exploration of the contralateral testis, in a survey performed in the United Kingdom and Ireland, 21 percent of the surgeons responded that they had not performed contralateral orchidopexy with concerns of damaging a healthy testis.[11] Conversely, Ahmed et al presented two cases with swelling of unilateral testis in newborns with normally appearing contralateral testis in whom bilateral testicular torsion was observed during surgical exploration. To the best of our knowledge, three case reports including six similar patients have been reported since 2008 (Table 1).[12-14,16-23] This finding also indicates the importance of exploration of the contralateral testis even with normal preoperative findings. This is especially important when the twisted testis has been removed.

Although sonography reported mild contralateral hydrocele, we explored the contralateral testis from a scrotal incision to prevent unwanted vasal or cord injury. Kaefer et al.[15] reported follow-up of 37 cases of neonatal testicular torsion with contralateral hydrocele that were explored with scrotal approach, in whom possibly patent processus vaginalis was present. They did not encounter any case of hernia recurrence during 1-14 years follow-up.

Full examination of the genitalia should be performed in each neonate. Bilateral exploration is mandatory in each case of perinatal torsion to evaluate the condition of contralateral testis and fix it to prevent future torsion that may end in anorchia.

**Informed Consent:** Written informed consent was obtained from parents of the boy for publication of this case report and relevant images.

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