Introduction
Neonatal testicular torsion is a rare event in which the cord of testis is twisted around its axis, leading to venous congestion and, if it continues, blood flow will gradually decrease and stop but, if not treated timely, it will bring about necrosis and dysfunction of the affected testis [1,2]. The torsion is mainly caused by the lack of repair in the testis and tunica vaginalis to the scrotal wall. In a study the prevalence of neonatal torsion is reported 6.1 in 100,000 newborn babies [3].

Case Report


Neonatal testicular torsion after cesarean section with spinal anesthesia: a presentation of two cases

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Abstract
Testicular torsion is a medical emergency appearing at any age, the prevalence of which in the neonatal period comprises 10% of the whole cases occurring at any point in life. Among all the cases being diagnosed at birth, 70% has occurred in the prenatal period and 30% in the neonatal period. Generally, the most common symptoms of testicular torsion during postnatal period include acute pain and swelling of the testis, but in the neonatal period it is often manifested by firmness and swelling of the testicle. Accordingly, careful attention of the physician and the medical staff while examining the newborn results in quick diagnosis and timely medical treatment of the torsion. This medical intervention prevents irreversible necrosis of the affected testis—the main consequence of the torsion—and helps to restore its function.

The major purpose of this study was to draw the attention of physicians and medical teams to prompt diagnosis of testicular torsion. This study presents two newborn babies with testicular torsion, diagnosed at early hours after delivery, who underwent surgical treatment within first 8-10 hours after birth.

Keywords
testicular torsion, orchiectomy, orchiopexy, spinal anesthesia

In their studies in 1990, Das and Singer reported testicular torsion as torsion of the spermatic cord and that of the appendicular testis. Torsion of the spermatic cord was classified into intravaginal and extravaginal; the latter is the most common form of neonatal testicular torsion (95%) and only 5% of the cases are intravaginal. Torsion of the left testis is more frequent compared to its manifestation on the right side [4,6].

In intravaginal testicular torsion, that usually reveals in adolescence [7], the testis undergoes bell clapper deformity and is situated horizontally in the scrotum, but in extravaginal torsion...
the entire cord twists about its axis. Spermatic cord torsion can be treated through prompt manual detorsion or urgent surgical operation.

The appendicular testis, a remaining part of müllerian duct, is a stalk-like structure attached to the upper pole of the testis. It appears in 90% of men and in 60% of the cases occurs bilaterally [2]. Torsion of the testis appendix is the most common cause of testicular torsion among 2 to 10-year-old boys and usually treated with rest, analgesic and anti-inflammatory agents (NSIDS), and the application of cold water pack to the testis area. It is difficult in some cases to differentiate between the torsion of spermatic cord and torsion of the testis appendix, and surgical intervention is necessary in suspicious cases.

Clinical symptoms of spermatic cord torsion in the neonatal period include firm and swollen testis, absent cremasteric reflex, undescended testis, and discolored scrotal skin (ecchymosis or blue dot sign under scrotal skin in the area of torsion) [5,6]. Acute sudden pain in the testis together with nausea and vomiting would be associated with these symptoms at higher ages. Although the newborn suffers from spermatic cord torsion, he is not agitated nor crying, therefore careful examination plays an essential role in timely diagnosis of the torsion.

If the acute testicular torsion is diagnosed at or soon after birth (within the first 4-6 hours) and an appropriate medical treatment is applied, the affected testis will not undergo irreversible necrosis and its function will usually be kept [4]. The treatment in golden time (within the first 4-6 hours) involves either detorsion of the affected testis by inducing local anesthesia without performing an open surgery or detorsion of the testis through an open surgery and fixing it to the scrotum. To avoid contralateral torsion, which may occur in the month following an open surgery, the testicle on the opposite side should be also fixed in the scrotum. If the golden time has passed and necrosis formation has emerged in the affected testis, surgical intervention is required to perform ipsilateral orchiectomy and contralateral orchiopexy. However, some authors believe that emergency surgical operation is not indicated in case of definite diagnosis of testicular torsion to have occurred during the prenatal period.

Predisposing factors of the neonatal testicular torsion are as follows [7,8,9]:
1. Embryonic asphyxia during pregnancy
2. Increased intrauterine pressure
3. Multiple pregnancy
4. Congenital disorders of tunica vaginalis and spermatic cord

The major predisposing factors of the testicular torsion after birth include:
1. Trauma
2. Sex hormones activities
3. Cold climate
4. Undescended testis
5. Strong cremasteric contractions
6. Exercise

Although careful physical examination and taking the clinical symptoms into consideration are the most important factors in diagnosing the torsion [12] these measures may facilitate the diagnostic procedure as well:
1. Color Doppler ultrasonography of the testis, spermatic cord and inguinal canal
2. Power Doppler ultrasonography
3. 99mTc-radioisotope scintigraphy

Case presentation
First Case
This study is a clinical evaluation of the two male neonates with testicular torsion, whose mothers underwent surgery with spinal anesthesia and they were both delivered by cesarean section in Lolagar Hospital-Tehran, in June and August, 2008, respectively.

The first was a full-term male newborn delivered in June 2008, whose mother, M.K., was 23, with GR=1. This mother had no history of taking drugs, medicinal allergy and disease before and during pregnancy. Based on the color Doppler ultrasonography in the 35th week of
pregnancy as well as chronic asphyxia. The intrauterine growth retardation (IUGR) was diagnosed as the indication for performing cesarean section. Below are the findings based on the patient's examination:

- T: 37.1°C
- BP: 110/75 mmHg
- HR: 90/min
- RR: 14/min

Class one airway and grade I to II laryngoscopy were evaluated in the airway exploration. Mucous membrane was not pale, and the motion of the jaw and neck was normal. The auscultation of heart sounds was S1-S2 and normal for the lungs. No abnormality was observed in the abdomen and limbs. Below are the laboratory tests results obtained before the patient had entered the operating room:

- WBC: 7600/mm³
  (Poly: 65%; Lymph: 32%; Eos: 2%; Baso: 1%)
- RBC: 5800000/mm³
- Hg: 12.3 gr/dl
- HCT: 38%
- Platelet: 160000/mm³

Since there was no contraindication to spinal anesthesia, it was chosen to be exerted for the patient. She was then transferred to the operating room. Prior to induction of spinal anesthesia, 500 ml of Ringer's solution was infused through an 18-gauge iv cannula inserted into a vein in her right elbow region for 20 minutes. Then she was placed in the sitting position and after prep and drep, through the L4-L5 intervertebral space, 70 ml of lidocaine 5% was injected into the subarachnoid space and she received necessary cares subsequent to the induction of anesthesia and the patient was moved into the supine position afterwards. All the necessary cares concerning spinal anesthesia during the operation were made.

The surgical exploration was performed and the neonate delivered with Apgar score 8 and 10 in one and five minutes, respectively. Vital signs of the patient remained stable during the operation, which lasted around 50 minutes. The volume of bleeding was about 850 ml and that of urine output was approximately 270 ml.

The newborn baby was examined by a pediatrician and the results were reported as follows:

- Height: 51 cm
- Weight: 3 kg
- Head circumference: 34 cm
- Chest circumference: 36 cm

Physical examination of the neonate was performed and no problem was found but the examination of genitals showed the left testicle was abnormally enlarged and pulled upward, with a firm solid mass, and the overlying skin was bluish without the presence of cremasteric reflex. The newborn was calm, without crying. Considering the suspected left testicular torsion, the color Doppler ultrasonography of the left testis, spermatic cord and inguinal canal were requested urgently. The report indicated the absence of flow of the left spermatic cord, under the torse area, left testicular atrophy associated with mild hydrocele, and a mass of about 7 mm in the spermatic cord, which may all resulted from the spermatic cord torsion.

The right inguinal canal and testicle appeared normal based on the ultrasonography. All the procedures were carried out within the first 6 hours and the newborn baby was referred, as an emergency, to a subspecialty center for pediatrics surgery. A pediatric surgeon visited him and the surgery was performed urgently. Since overwhelming atrophy was found in the left testicle, the ipsilateral orchiectomy and contralateral orchiopexy were performed. Nonetheless pathologic examination of the affected testis verified total atrophy and chronic necrosis of the left testis and spermatic cord beneath the torse testicle. The clinical examination, direct observation of the torsion during the surgery, histopathologic examination and the color Doppler ultrasonography identified a left testicu-
lar torsion of both intrauterine and extravaginal type.

**Second Case**

The second newborn boy was delivered in August 2008, whose mother, F.A., 32, GR=2 and full-term pregnancy, with an indication of repeated cesarean section, fetal distress and meconium stain of amniotic fluid was a candidate for cesarean delivery. She had no history of drug intake, drug allergy, and disease before and during pregnancy. She had undergone cesarean section four years ago under general anesthesia without any complication. Below are her report of laboratory tests taken before she was admitted to the hospital:

- **WBC:** 8200/mm³
  - (Poly: 67%; Lymph: 30%; Eos: 2%; Baso: 1%)
- **RBC:** 4100000/mm³
- **Hg:** 11.8 gr/dl
- **Hct:** 35%
- **Platelet:** 119000/mm³

Physical examination of this mother showed no specific problem including anesthesia-related ones. Since no contraindication was found to the spinal anesthesia, it was chosen as a safe anesthetic technique for the patient. Her vital signs prior to the induction of spinal anesthesia were as follows:

- **T:** 36.8°C
- **BP:** 117/80 mmHg
- **HR:** 78/min
- **RR:** 12/min

An 18-gauge iv cannula was inserted into a vein in her right elbow region and 500 ml of Ringer's solution was infused within half an hour. She was then placed in the sitting position and after prep and drep, considering the patient's condition, 60 ml of lidocaine 5% was injected through the L4-L5 intervertebral space. Routine cares concerning spinal anesthesia induction were provided, accordingly.

The operation was performed and the baby delivered. The volume of bleeding during the surgery, which took about an hour, was 1000 ml and the urine output was 330 ml. Regarding the conditions of surgery and patient, 2000 ml of D5NS and 1000 ml of Ringer's solution were infused during the operation. The neonate's one-minute and five-minute Apgar scores were 8 and 10, respectively. He was examined by a pediatrician and the following results were found:

- **Height:** 55 cm
- **Weight:** 4.250 kg
- **Head circumference:** 32.5 cm
- **Chest circumference:** 36

Physical examination of the neonate revealed no particular problem but urogenital examination manifested the right testicle as hard and firm, and through transillumination, the scrotum looked swollen with fluid. Scrotal discoloration was not observed. The cremasteric reflex was absent and the affected testis was pulled upward. Nothing unusual was noted in physical examination of the left testicle. An urgent request for urological consultation was asked, and the color Doppler ultrasonography of the testes, spermatic cord and inguinal canal showed hydrocele in the right testis, absence of arterial flow in the right spermatic cord under the torsed area, and swelling as well as atrophy of the right testicle. The inguinal canal bilateral examination showed no abnormality, and the left testis looked normal.

Considering the findings above, the newborn baby was urgently referred to a pediatric surgery center and operated by a pediatric surgeon 8 hours after birth. The orchiectomy of the right testicle and contralateral orchiopexy were performed based on the sonography report and the surgeon's observations during the operation, which proved total atrophy of the right testis. The pathological report revealed atrophy and chronic necrosis of the right testis and spermatic cord. The testicular torsion was diagnosed to
have developed during the prenatal period and occurred extravaginally based on the clinical conditions of the neonate and reports of pathology and the Doppler ultrasonography. After 4 days hospitalization, the newborn was finally discharged with no encountered problems.

**Discussion**

Neonatal testicular torsion is a rare event [1,11,12] caused by inadequate fixation of the testicle within the scrotum and, consequently, its excessive movements lead to the torsion. The symptoms of the torsion emerge as a result of the arterial occlusion of the testis and, eventually, necrosis and atrophy of the testicle [10]. The testicular torsion is generally classified into two categories:[1,2,3]

1. torsion of the spermatic cord
2. torsion of the appendicular testis

Torsion of the spermatic cord may occur either extravaginally or intravaginally. Extravaginal torsion involves the spermatic cord in which the cord twists around its axis and if it persists it will give rise to the total obstruction of blood flow and necrosis as well as dysfunction of the organ finally. It is considered as a medical emergency and should be treated within 4-6 hours, after which the treatment will not be usually effective and orchiectomy of the affected testis must be performed. According to most of the researchers, since the contralateral testicular torsion is highly likely in the next month, the contralateral testicle should be fixed within the scrotum.

Torsion of the appendicular testis is usually prevalent among boys of 2-to 10-year old, resulting from trauma, hormonal stimulation, exercise, cold environment and strong cremasteric contraction. Usually it does not require any emergency surgical intervention and can be treated conservatively, including rest, analgesic and NSAIDS agents. If diagnosis is suspected whether the torsion has involved the appendicular testis or the spermatic cord, prompt surgical exploration and treatment is indicated.

Neonatal testicular torsion comprises 10% of all testicular torsions, which mostly occurs extravaginally. In 70% of the cases it occurs in the prenatal period and the rest (30%) takes place after birth or during neonatal period. Timely diagnosis of the neonatal testicular torsion is of immense importance since it is often manifested as an enlarged testis, and the newborn does not cry and is not agitated. A careful physical examination can help detect a swelling in the area of the spermatic cord torsion, absent cremasteric reflex and undescended torsion. However, the classic manifestations of acute torsion at older ages include pain and discomfort, and the patient helps the physician locate the painful area which contributes to diagnosis and follow-up of the disease.

To diagnose the torsion, helpful paraclinical procedures generally consist of color Doppler, power Doppler ultrasonography and 99mTc - radioisotope scintigraphy. Taking the torsion into account and careful clinical examination play prominent roles in diagnosis, however. (The procedures are employed to detect arterial flow and perfusion of the spermatic cord and the testis.)

Both of the patients studied suffered from neonatal testicular torsion of extravaginal type, the prevalence of which in the prenatal period has been reported 70%. The torsion had involved the left testis and right testis in the first and second cases, respectively; reliable sources12 have reported more left-sided testicular torsions than the right-sided.

Based on the Doppler ultrasonography of both newborns, intrauterine growth retardation (IUGR) and fetal distress were considered the possible causes of the torsion in the first and second newborn babies in the prenatal period, respectively; they are both introduced as the predisposing causes of the neonatal testicular torsion by the reliable sources.

As in both babies the testicular torsion had occurred during the prenatal period and testicular atrophy was observed in both cases, the or-
chidectomy of the affected testis and the contralateral orchiopexy were performed; this procedure is also in accordance with the reliable urological and pediatric surgery sources [2,11].

In both neonates, clinical examination revealed swelling and firmness of the affected testicle accompanied by a high-riding testis, an absent cremasteric reflex and hydrocele; they are introduced as the major symptoms of the neonatal testicular torsion in the reliable sources2 as well.

Crying and discomfort were not observed in either neonates; they are not mentioned as the main symptoms of neonatal testicular torsion in the urological and pediatric surgery sources2, also.

Discoloration of the scrotal skin (ecchymosis) and hydrocele were present in one of the babies and absent in the other; in the reliable sources discoloration of the scrotal skin is also not reported for all the testicular torsions.

Testicular torsion in both newborns was unilateral; in reliable urological sources1,11 90% and 10% of the cases reported are unilateral and bilateral, respectively.

Surgical operation in both cases had been performed 7-8 hours after their birth, exceeding the golden time.5 (The golden time for surgical intervention in testicular torsion is often 4-6 hours after birth, although, in both cases, the testicular torsion had occurred during prenatal period and atrophy of the testis had already taken place.)

Prior to the surgical operation a color Doppler ultrasonography was performed, in both cases, which revealed absent arterial flow of cremasteric cord in affected testis, and atrophy of the torsed testicle associated with hydrocele.

Mothers of both newborns had undergone cesarean section with spinal anesthesia; no relation between delivery method and testicular torsion was reported by reliable sources.

The pathology report of the testicle and spermatic cord, taken during the surgery, confirmed chronic necrosis and total atrophy of the affected testis during the prenatal period.

Result

Neonatal testicular torsion is considered as a medical emergency, and the affected testis will be avoided necrosis and dysfunction if careful medical examination and timely treatment procedure are performed. Since the neonate is unable to express the symptoms of the disease in the neonatal period, among which agitation and crying are frequent symptoms of an acute testicular torsion, vigilant attention of the physician and medical staff and careful clinical examination may prevent neonatal testicular torsion.

References


