Abstract

Neonatal testicular torsion, also known as perinatal testicular torsion is a subject of debate among surgeons. Neonatal testicular torsion either intraperitoneal or postnatal results into extravascular torsion which is a different entity than intravaginal type but has the same devastating consequences if not diagnosed and managed well in time. Testicular torsion results into acute ischemia with its resultant sequelae such as abnormality of testicular function and fertility. Urgent surgical exploration and fixation of the other testis are the key points in the management. General anesthesia is not a contraindication for exploration as thought before. Diagnosis and controversies on management of testicular torsion are discussed in this review.

Key Words: Testis; Testicle; Testicular Torsion; Neonatal; Perinatal

Introduction

Testicular torsion was first described in 1840 by Delasaiuvel[1]. The condition was first time reported in the newborn by Taylor in 1897[2]. But it has taken a long time for it to be recognized as a vascular emergency that needs prompt diagnosis and urgent exploration. It was not until 1907 when Rigby and Howard wrote their classic paper on torsion that this entity gained widespread clinical acceptance[3]. Neonatal testicular torsion also known as perinatal testicular torsion (PTT) is a rare event with controversies regarding its etiology, presentation, surgical management and sequelae. Initially, PTT was thought of one distinct entity. Recently it has been subcategorized as either occurring prenatally in utero or postnatally in the first month of life[4].

Prenatal torsion is marked by minimal to no discomfort and few localized findings. Conversely, postnatal torsion is an acute manifestation with considerable tenderness and swelling of a previously normal testicle[5].

A review of the urological literature demonstrates no consistent pattern regarding the potential etiologies of PTT. Possible theories include difficult labor, breech presentation, high birth weight, an over reactive cremasteric reflex and multiparity[5,6]. Despite the type or mechanism of torsion, PTT is an unexpected finding for neonatologists and parents. Antenatal ultrasound is not sensitive in detecting this abnormality; thus, it is commonly diagnosed during the routine postnatal physical examination. The pediatric urologist or pediatric surgeon is usually consulted in the first few hours of life and is faced with formulating a management plan. There is much controversy regarding the optimal management.

In this article, we will try to elaborate different options of management and review of literature.
Embryology and Anatomy

Shortly after 6 weeks' gestation, the testis-determining SRY gene on chromosome Y directly affects the differentiation of the indifferent gonad into a testis. Around 6-7 weeks' gestation, Sertoli cells develop and secrete Müllerian inhibitory substance (MIS), which leads to the regression of the female genital organs.

Around the 9 weeks' gestation, Leydig cells start producing testosterone, which promotes development of the Wolffian duct into portions of the male genital tract. Because of the differential growth of the fetus, the testicles move into the pelvis, close to the internal ring. But initially it develops retroperitoneally adjacent to the kidney[8]. At about the third month of intrauterine life, the gubernaculum testis develops and extends from the genital tubercle to the inferior pole of the testis via the inguinal canal. The peritoneum later encircles the testis completely forming a mesentery, the mesorchiwm. The inferior main part of the gubernaculum attaches to the scrotal skin pouch and the minor, superior part disappears[8].

The testes are paired structures suspended in the scrotum by the spermatic cord. They measure approximately 2.0 × 3.0 × 4.0 cm in the adult and weigh 15-20 g, but the measures are variable in the neonate.

Each testis is surrounded by a fibrous capsule called the tunica albuginea. The testis contains seminiferous tubules, which are tightly coiled and arranged in wedge shaped lobules. The margins of the lobules are formed by septations arising from inner layers of the tunica albuginea. The seminiferous tubules converge toward the mediastinum testis and unite to form larger tubules, which in turn form a network of tubules, the rete testes. The rete testes unite to form efferent tubules at the superior pole of the testes. These efferent tubules (10-15 in number) constitute the head of the epididymis. The efferent tubules then unite to form a single lumen structure called ductus epididymis (Fig. 1).

The ductus epididymis forms the body and the tail of the epididymis, which is located on the posterolateral aspect of the testis. The tail of the epididymis undertakes an acute turn and continues as the vas deferens, which joins the spermatic cord. The testis and epididymis are vested by an extension of the peritoneum called the tunica vaginalis; this covers all but the posterior side (Fig. 1).

The testis is suspended from the spermatic cord like an object at the end of a rope. The contents of the spermatic cord include the vas deferens and its artery, the testicular artery, the pampiniform plexus of veins, lymphatic vessels, and the sympathetic nerves. All of the aforementioned structures are enclosed in facial layers derived from the oblique muscles of the abdomen. Loops of cremasteric muscle encircle the spermatic cord and scrotum, innervated by the ilioinguinal nerve and are responsible for the cremasteric reflex[8].

The blood supply to the testis is mainly from the testicular artery, a branch of the abdominal aorta. Contribution is also made by the deferential artery,
a branch of the hypogastric or superior vesical artery; it primarily supplies the vas deferens. In addition, the cremasteric branch of the inferior epigastric artery forms a network over the tunica and forms an anastomosis at the testicular mediastinum. The testicular artery enters the testis through the mediastinum and branches under the tunica albuginea to form capsular arteries. The capsular arteries send radial branches, known as the centripetal arteries, into the substance of the testis. The centripetal arteries form U loops near their ends, increasing the effective area of supply. The scrotal wall is supplied by the pudendal artery, which is not a content of the spermatic cord and thus not involved in testicular torsion (Fig. 2).

The testicular veins exit from the mediastinum and form a plexus called the pampiniform plexus. The plexus then combines into a dominant vein, which follows the testicular artery into the spermatic cord. The left testicular vein drains into the left renal vein, whereas the right testicular vein enters directly into the inferior vena cava.

**Pathology and Pathogenesis**

Two types of testicular torsion are recognized, and each has slightly different etiologies. Extravaginal torsion occurs in fetuses and in neonates. With this type of testicular torsion, the testis, epididymis, and tunica vaginalis twist on the spermatic cord[9] (Fig. 3a,4). In result of twisting ischemic changes such as swelling, degeneration, necrosis and infarction occur. Torsion is usually away from the midline due to the orientation of cremasteric muscle fibers. The degree of torsion varies from 180° to more than 720°. The severity of torsion depends upon the degree of twist. In result of torsion, both venous and arterial blood supplies are hindered and ultimately results into gangrene of the testis. PTT may be unilateral or bilateral, and bilateral torsion can be synchronous or asynchronous. Baglaj M et al mentioned incidence of bilateral synchronous torsion as 67% and asynchronous 33%[10]. Intravaginal torsion occurs more commonly in the peripubertal period than at other times. This type of testicular torsion is associated with a bell-clapper deformity (Fig 3b).

**Perinatal History**

In case of perinatal testicular torsion, prenatal history is very important. It should include pre eclampsia, gestational diabetes, twin gestation, large size for gestational age, presence of prenatal hydronephrosis. Birth history should include mode of delivery, as well as presence or absence of nuchal cord, meconium aspiration and prolonged

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Fig. 2a: Arterial supply of testis and epididymis

1) Testicular artery
2) Deferential artery
3) Cremasteric artery

2b: Transverse section of the testis

4)Internal spermatic fascia
5)Parietal and visceral lamina of tunica vaginalis
6)Testicular artery
7)Capsular arteries
8)Centripetal arteries
9)Vas deference
delivery. Jonathan D Kaye described 10 patients in whom prenatal history was known, 5 (50%) had at least one significant prenatal finding. Two mothers had preeclampsia, two mothers had gestational diabetes, 1 neonate was large for gestational age and 1 neonate was diagnosed prenatally with hydronephrosis. Two patients were product of twin gestation. 5 of 10 patients were delivered with some sort of delivery complication[11].

While postnatal torsion has not been linked to any specific risk factors, such is not the case for prenatal torsion. This condition has been associated with breach presentation and traumatic delivery. Most prenatal torsions occur in full term neonates with the mean birth weight in one series being 3.6 kg[12]. All of these factors can potentially increase intrauterine pressure as well as pressure in the birth canal during parturition[13]. It is plausible that such pressure may, in turn, stimulate a brisk cremasteric response in the setting of loose tunic scrotal attachment[14]. So a detailed prenatal history and mode of delivery are mandatory and certainly in postnatal torsion history of initial well-being, irritability, vomiting, and sudden scrotal swelling should also be considered.

**Clinical Findings**

After obtaining a detailed history, examination of the spine, back, perineum, groin and the scrotum should be performed. Examination of the abdomen is also important to exclude other problems giving rise to such symptomatology. On physical examination, the affected hemiscrotum is swollen and frequently erythematous. The normal separation of the testis from the epididymis may not be palpable. An elevated, horizontal lie of the affected testis and skin pitting at the scrotal base may provide evidence in support of the diagnosis. With transillumination, the ischemic testicle may be visualized (*blue-dot sign*). Typically, no pain relief occurs with scrotal elevation, as observed with acute epididymitis. The spermatic cord is typically thickened and tender. Tenderness alone may be indicative of acute epididymitis. In a practical sense, PTT includes neonates with 5 definite clinical pictures[15].

1) If torsion occurs in prenatal period far from birth, the newborn will be born with an absent or a nubbin testis. In the first case (*vanishing testis*), the patient should be treated as cryptorchid at an older age. In the second case he should be approached as a sequel of a long standing intrauterine testicular torsion.

![Fig. 3: Types of testis torsion](image)

- a) Extra-vaginal
- b) Intra-vaginal (Bell-Clapper Deformity)
2) If torsion occurs in the prenatal period far from birth, the newborn will be born with an absent or a nubbin testis. In the first case (vanishing testis), the patient should be treated as cryptorchid at an older age. In the second case he should be approached as a sequel of a long standing intrauterine testicular torsion.

3) If torsion occurs in the prenatal period far from birth (several weeks), typically the child will present since birth a regular, firm, painless scrotal mass, often in the upper part of the hemiscrotum, smaller than the contralateral normal testis, very attached to the scrotal wall, without acute inflammatory signs, and which does not transmit light.

4) If torsion occurs in the prenatal period near birth (several days), the newborn will present since birth a firm and painless scrotal mass, bigger or similar in size than the contralateral normal testis, without acute inflammatory signs and which does not transmit light.

5) If torsion occurs in the prenatal period very near to birth (few days or several hours), the newborn will be born with acute inflammatory scrotal signs: a painful, enlarged, bluish or reddish hemiscrotum with an enlarged and sometimes elevated testis that does not transmit light, and a thickened and painful cord.

6) If torsion occurs in postnatal period within the first month of life, the child will be born without any scrotal signs (occasionally a hydrocele) and the acute inflammatory signs will appear later.

**WORK UP**

In addition to careful clinical examination and evaluation, Doppler sonography is increasingly used in the management of patients with suspicion for testicular torsion[16-18]. This can mainly be attributed to the quick technical progress with excellent anatomical imaging and simultaneous portrayal of blood flow[19]. High frequency transducers, power Doppler and tissue harmonic functions are increasingly used in daily routine care. However to prove testicular torsion, surgical exploration of the scrotum is still fairly often performed. Taken into consideration that less than 30% of the infants with an acute scrotum suffer from testicular torsion[17], it is obvious that most infants may be managed conservatively. Despite reports on strongly varying results concerning the reliability for detecting testicular torsion by Doppler sonography[20], recent studies show an increasing sensitivity of this technique[21,22].

In neonates color Doppler sonography (CDU) shows an enlarged, heterogenous testis, thickened tunica albuginea with rim like hyperechoic reflections (calcifications) at the transitional zone between testis and tunica albuginea. Hypoechoic central area may also be evident which shows necrosis[23]. In neonatal torsion CDU may be used to estimate the time elapsed since the occurrence of intrauterine testis[24]. A relatively short duration of torsion is characterized by mixed echogenecity. Prolonged intrauterine torsion shows calcification and a hypervascular ring of tunica with a hypodense center[24]. While performing Doppler ultrasound, description of the

*Fig. 4: Testis torsion. a: Erythema of the scrotum over torted testis (presented on 4th day of life with acute right hemiscrotum for 12 hrs), b: Intraoperative photo showing torted gangrenous testis*
echogenicity and tissue structure of the testes and epididymis of both testes, scrotum volumetry inside comparison and assessment of central and peripheral blood flow of the testicles at power and CDU must be considered. In case of given perfusion, the resistance index (RI) should be included to recognize possible partial torsion[25].

Special attention has to be paid to the fact that spontaneous reduction in intermittent torsion can appear as reactive hyperperfusion of the testicular parenchyma. Additionally, one should take into consideration that in case of testicular torsion with absent central blood flow, peripheral perfusion can be maintained via collateral arterial supply. Pulsed Doppler sonography with mechanical sector scanners is a better method than CDU for the diagnosis of testicular torsion[26]. But this technique studies only the testicular arteries and omits the scrotal, paratesticular and testicular surface vessels.

Radionuclear scanning is another diagnostic modality in neonatal testicular torsion. Technetium-99m pertechnetate is the agent of choice, with a pediatric dose of at least 5 mCi. Typically, immediate radionuclide angiograms are obtained, with subsequent static images as well. In the healthy patient, images show symmetric flow to the testes, and delayed images show uniformly symmetric activity. The appearance of testicular torsion on scintigraphy depends upon the chronicity. In acute torsion (usually <7 h), blood flow may range from normal to absent on the involved side, and a nubbin sign may be visible. The nubbin sign is a focal medial projection from the iliac artery representing reactive increased flow in the spermatic cord vessels terminating at the site of torsion (This sign can also be seen in later stages). Static images demonstrate a photopenic area in the involved testis. In the subacute and late phases of torsion (missed torsion), there is often increased flow to the affected hemiscrotum via the pudendal artery with a photopenic testis and a rim of surrounding increased activity on static images. This has been called a rim, doughnut, or bull's eye sign.

Acute epididymitis generally appears as an area of focal or diffuse increased activity in the involved hemiscrotum. Testicular appendix torsion has a variable appearance: it may have a normal scan or a focal area of increased or decreased activity. An abscess, tumor, or hematoma may be indistinguishable from a torted testicle, demonstrating a hyperemic rim surrounding an area of decreased activity. Some studies have claimed it to be a better diagnostic modality than CDU[27].

**Management**

Perinatal testicular torsion (intrauterine and postnatal in the first 30 days) is an uncommon entity and represents about 12% of all testicular torsions during infancy[28]. It is thought that the majority (70%) of perinatal torsions are present at delivery and 30% develop postnatally in the first month of life[29]. Review of literature indicates that there is controversial management of this entity. We have tried to review the literature on optimum management of intrauterine and postnatal testicular torsion and controversies whether contralateral testis should be fixed or not. We have also reviewed the literature on methods of testicular fixation.

Jonathan D. Kaye et al[11] wrote their recommendation in the light of their experience on 15 cases of neonatal testicular torsion. According to them in cases of bilateral testicular torsion emergent bilateral exploration via inguinal approach should be performed and the surgeon should err towards orchidectomy rather than orchiectomy. If findings are present at birth and torsion is unilateral, they recommend ipsilateral orchiectomy and contralateral orchidopexy (both via inguinal approach) after 1 month of age with interim parental scrotal examination. If findings are not present at birth (i.e. postnatal torsion), they advised emergent ipsilateral exploration and contralateral dartos pouch orchidopexy (both via inguinal approach).

Jose L Cuervo et al managed their patients by considering whether the torsion was long standing intrauterine, very near delivery or postnatal. For long standing intrauterine torsion they recommended that as there is no urgency these neonates should be operated on electively when the child is in optimal clinical status to confirm the suspected diagnosis, 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 and one should not spend valuable time in ultrasound studies[15].

Many investigators have recommended that boys who present with suspected unilateral torsion within the first 30 days of age, the risks associated with early surgery and anesthesia outweigh the prospect of salvaging the testis[6]. It is now clear that the potential untoward anesthetic risks involving an otherwise healthy neonate quoted in prior studies are outdated and are not consistent with the current standards of pediatric anesthetic practice[30]. Those opposed to emergent surgical exploration of a suspected torted testis often cite the experience of Kaplan and Silber[31], who reported a salvage rate of only 5% in their experience with babies who presented with testicular torsion. However, this percentage included all torted testes presenting in neonates. It ignored the dramatic differences between an acute torsion presenting after birth and the more common entity of the long standing event of antenatal testicular torsion. A subsequent survey conducted by Das and Singer[29] suggested that in boys with neonatal torsion, at least 28% occurred postnatally. This suggests a potentially greater rate of testicular salvage than the disappointing results previously reported. Following a policy of managing postnatally diagnosed torsion in all neonates in a fashion identical to that practiced in older boys, Pinto et al[32] were able to salvage 2 (20%) of 10 testes with emergent surgical exploration.

Al-Salem [33], Guiney et al[12] and Logino et al[34] adopted the policy of early surgical intervention. John et al[35] reviewed the literature on neonatal testicular torsion. They described 77 boys treated for neonatal torsion, no testes were salvaged. This dismal outcome underlines that immediate surgical exploration, although commonly performed may not be saving torted testes.

Baglaj M and Carachi R[10] mentioned their experience of 58 neonates with neonatal testicular torsion. They strongly recommend an emergency operation in all males presenting with clinical symptoms of unilateral or bilateral testicular torsion. They believe that prompt exploration serves its use as the most objective diagnostic method. They are of the opinion that clinical judgement by a pediatric surgeon or urologist is of utmost importance not the ultrasound. Although the prognosis for these patients is poor, an elective delay in operative intervention seems to be inappropriate from medical and medicolegal point of view. Taking into consideration a risk, however small of asynchronous torsion, bilateral emergent exploration is strongly advised in all neonates.

Yerkes EB et al[36], are also in favor of immediate surgical exploration. According to them surgery is the best method available to confirm testicular non viability and small but present risk of asynchronous torsion in the perinatal period can be addressed early by pexing the contralateral testis. They also mentioned that perinatal torsion is asymptomatic and in its early stages may show no clinical signs of having occurred, thus early surgical exploration may detect asynchronous torsion and allow its correction.

Another controversial issue is whether contralateral orchidopexy is justified. Some investigators suggested that since predisposing factors are lacking in extravaginal torsion, there is no need for contralateral orchidopexy[37]. On the other hand the increasing number of reported cases with bilateral intrauterine torsion supports a predisposing factor[38-40], although asynchronous bilateral torsion is rare, it can, however occur at any time and has been reported as early as 48 hours after torsion on the other side[39]. Kashif and Riazulhaq et al mentioned 11 cases of neonatal testicular torsion, opposite testis was fixed in all cases[41]. Mishriki et al are also of the opinion to fix a single testis whatever the cause is[42]. So there is consensus in exploring the contralateral side in the same operation or soon after that, depending upon the local conditions and clinical status of the patient[4,29,30,32,33,39,41,42].

But no consensus exists on how such fixation should be achieved. There is wide variation in the practice of pediatric surgeons, which may reflect that all techniques are equally efficacious at pre-empting recurrence, although some methods purposely avoid breaching the integrity of the tunica albuginea. Suture fixation has been described with both absorbable[43] and non absorbable sutures[44]. But concerns regarding testicular damage due to needle trauma have led some to adopt sutureless fixation methods, such as Jaboulay procedure[45] or creation of a dartos pouch[46]. Simp.e suture fixation results only in fine adhesions at the suture points[47] but when a window in the tunica vaginalis was created, dense adhesions between the tunica vaginalis and the
scrotal wall were seen. Literature review of recurrent torsion after previous fixation has identified 20 cases in one study[48]. In 15 of 17 cases where the type of suture at the original repair was specified, an absorbable suture had been used. Two cases of recurrence after fixation with non absorbable sutures were also reported. Bolln C et al[49] wrote their results on operative management of testicular torsion and according to them 16 of the 95 surgeons in their study had experience with torsion after previous fixation and the recurrence was associated with the use of absorbable sutures in all instances. They concluded that method of fixation remains a matter of personal preference but the use of absorbable sutures for suture fixation is associated with recurrence of torsion, and should be avoided.

**Conclusion**

Management of perinatal testicular torsion is a matter of controversy. Although the possibility of salvaging the involved testicle is very low, however, it is hard to justify a passive approach to a condition resulting in such a devastating condition as anorchia. Although risk of general anesthesia can not be excluded 100 percent, but in modern era it is not a contraindication. Time should not be wasted in doing investigations because imaging studies have a limited role in preoperative management. One should explore the affected side promptly to confirm the diagnosis and to fix or remove the affected testicle. The contralateral scrotum also should be explored because of the risk of asynchronous contralateral testicular torsion. Whatever method of fixation is used, non absorbable suture is advisable.

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