PERINATAL TESTICULAR TORSION
Testicular torsion during fetal and neonatal period

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ABSTRACT: The term perinatal testicular torsion (PTT) defines the extravaginal testicular torsion that happens either during the fetal period (prenatal testicular torsion) or the neonatal period (postnatal testicular torsion). It concerns 10-22% of testicular torsions that appear during childhood. The clinical findings of PTT after birth depend on the time of the event. In every newborn male with scrotal swelling and discoloration, while the other testis is hard, PTT should be considered as the diagnosis until proven otherwise. The treatment of PTT is discussed by the authors. Disagreement is present concerning the time and method of treatment, as well as the necessity of preventing orchidopexy of the contralateral testis. In our study, we review recent literature in order to establish an evidence based conclusion over treatment.

Keywords: Perinatal testicular torsion, extravaginal torsion, scrotal swelling, vanishing testis syndrome

DEFINITION AND EPIDEMIOLOGY

By the international term perinatal testicular torsion (PTT) is defined the torsion that occurs between the fetal period and the first 30 days of life. The incidence of PTT is 6,1/100.000 male neonates and concerns 10-22% of testicular torsion's episodes during childhood (1,2,3). 70-81% of all the cases happen during the fetal period (prenatal testicular torsion) and 19-30% during the neonatal period (postnatal testicular torsion) (3,4). At 30-48% of the cases the torsion concerns the left testis, at 44-55% the right while 8-15% is bilateral. (3,5). Kaye J, Levitt SB et al in their study described 16 cases of PTT and found that the degree of the spermatic cord's torsion ranged from 360 to 1440 degrees (3). Generally, at 5-20% of all cases torsion can be bilateral at the same or different time. (6,7,8). At an eventual bilateral torsion it is remarkable that clinical signs can be unilateral (9). Experimental studies have proven that testicular torsion can cause irreversible damage of spermatogenesis after 4-6 hours of ischemia. A prolonged ischemia of 10-12 hours can cause an irreversible damage at the endocrine function of the testis. (10,11,12)

HISTORIC BACKGROUND

Testicular torsion was firstly described in 1840 by Delasiauve (13). In 1897 Taylor was the first who described the clinical signs of testicular torsion at a newborn male, but without determining this entity as a vascular event (14). Rigby and Howard published in 1907 the first complete study over neonatal testicular torsion (15). Papadatos C and Moutsouris C published in 1967 a case of bilateral PTT (16). Only 268 cases of prenatal testicular torsion have been described till now (17,18).

ETIOLOGY

PTT is mostly extravaginal (3,17,19,20). In a review study of Nandi B and Murphy FL it is concluded that 90,6% of PTT cases are extravaginal (5). Extravaginal torsion may include the cremaster muscle and all the layers of the spermatic cord (extravaginal torsion within dartos muscle) or can take place inside the internal spermatic fascia without the participation of the cremaster muscle. It is believed that a very important factor is the increase of the intra-abdominal pressure and indirectly to the inguinal channel, during labor (21). This

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leads to a contraction of the cremaster muscle followed by an abnormal movement of the testis due to weak adhesions of the gubernaculum to the scrotum (20,22). We must notice that, during perinatal period, the external spermatic fascia is not strongly attached with dartos, a fact that can explain the extravaginal torsion inside the cremaster muscle (23). The evolution of the extravaginal torsion inside the internal spermatic fascia could be attributed to the existence of weak adhesions between the parietal layer of tunica vaginalis and the external scrotal layers. This observation is not fully accepted because it is known that the testis is attached to the parietal layer of tunica vaginalis with wide mesorchium.

There is no clear cause of PTT. Factors that have been related with prenatal testicular torsion is preeclampsia, diabetes, multiple gestations, congenital hydronephrosis, higher birth weight, vaginal delivery, especially a difficult one, as well as meconium aspiration (17,18,24,25). In an older study, Guiney E. and Mc Glinchey concluded that PTT considered mostly neonates with birth weight over 3600g (26). On the other hand, no factors responsible for testicular torsion during the neonatal period have been found. (25)

CLINICAL PICTURE

Clinical signs of PTT can be estimated after birth and depend on the time of the event(18,19)

If it takes place before 26th week of gestation(WG) it occurs either a vanishing or a nubbin testis. Generally 2,9% of all cryptorchidism's cases are part of the vanishing testis syndrome. By the term vanishing testis we mean the presence of a fibrous, non –vascularised, nonfunctional testicular tissue. The fact that a vanishing testis is a result of a vascular event is proven by the histological findings, which include a hemorrhagic infraction and the presence of haemosiderin inside the macrophages of the specimen. (27,28,29,30). In the study of Broderick KM , Martin BG, Hemdon CDA et al , 69% of pediatric urologists believe that the blind ending of testicular vessels and spermatic cord -as we can see at the laparoscopic investigation of a non-palpable testis- correspond to the term vanishing testis syndrome due to a torsion in-utero. (31)

If PTT takes time between 26th and 32nd WG, then it results in a small, atrophic testis in the ipsilateral hemi-scrotum or its upper part. It appears like a spherical, painless, hard mass adhered to the scrotum, with no signs of inflammation. Light does not come through during transillumination.

If PTT appears 1-2 weeks before birth then the testis preserves its size, but is hard and painless with no signs of inflammation. Light does not come through during transillumination.

If PTT takes time among 5 days to few hours before birth the signs are those of an acute scrotum. The testis is painful at palpation, hard and swollen. Moreover, there is edema and sensitivity at the spermatic cord. The testis may be inside the scrotum or higher along its normal route, while the hemi - scrotum is bluish or reddish. Despite the appearance of a hydrocele in the tunica vaginalis, it does not transmit light during transillumination.

If PTT develops during the neonatal period then the scrotum appears normal at birth. In this case there is an increased possibility of torsion of the contralateral testis within the first 2 months of life (19).

DIAGNOSTIC APPROACH

In every male neonate with voluminous dark or reddish scrotum and reduced light passage at transillumination, in whom the ipsilateral testis is painful and hard at palpation, PTT is a probable diagnosis (25). Usually, the neonate does not seem to suffer and no systematic inflammatory process is observed.

Differential diagnosis of unilateral solid scrotal swelling includes incarcerated hernia, epididymitis, meconium periorchitis, scrotal hematoma due to birth injury , scrotal abscess ,ectopic spleen, ectopic adrenal and finally some rare tumors such as juvenile granulose cells tumor and yolk sac tumor. (17,32,33)

After the recognization of clinical signs of an acute scrotum we must proceed carrying out color doppler or better pulsed doppler sonography (34). Resistance index can contribute to the evaluation of arterial flow at a partial or a loose testicular torsion (35). In the case of a transient torsion, increased blood flow and homogeneity of testicular parenchyma should be evaluated (25). Absence of arterial flow in the center of the testis with preservation of peripheral blood flow can be induced by collateral testicular vascularisation during the torsion.

If the testis is necrotic, the imaging study shows swelling and thickening of visceral lamina of tunica vaginalis , hyperechoic réflexions in the periphery of the testis due to calcification, while the central part is partially hypoechoic. (36)

Technetium-99m pertechnate contributes to the diagnosis of acute scrotum if there is no indication for emergent operative exploration.

If the torsion occured before weeks or months, increased blood flow in the hemi-scrotum is depicted due to the increased vascularisation through the pudental artery as well as rim sign and bull's eye sign. This pathognomonic sign in static images is shown due to the photopenic central area (interruption of vascularisation) and the increased collateral peripheral circulation. (37)

After a recent torsion (before 6-10 hours) presence or abcense of vascular flow can be imaged. In case of absence, nubbin sign is recognized. This corresponds to the discontinuance of arterial flow originated from the internal iliac artery to the torsion area. Static images can show the characteristic photopenic area of the testis. (24)

TREATMENT

There is no agreement among the authors about the treatment of PTT. There are different opinions concerning the time and method of treatment, the necessity of preventive orchidopexy of the contralateral testis and the surgical removal of the non-viable testis.

In their study, Broderic KM, Martin BG et al (31) analyse 121 answers to the questionary they sent to the members of the Society for Fetal Urology and the Society for Pediatric Urology, as they are noted in table 1.

Concerning prenatal testicular torsion (PrTT), 34% would perform immediate investigation of the affected hemi-scrotum, 26% an urgent investigation, 28% a selective one the next 72 hours, while 12% would not interfere surgically. 93% would perform a contralateral orchidopexy. In case of PrTT an important reason for surgical investigation, besides the establishment of torsion's diagnosis and exclusion of other causes such as scrotal hematoma or tumor, is the the performance of a preventive orchidopexy of the unique contralateral testis. That testis can be tortued at the same time or after few days or weeks without a typical clinical image (subclinical torsion) (38). In case of bilateral PrTT 90% would perform an immediate investigation, 1% an urgent one, 2% in a selective base and 7% would not investigate at all.

When PTT takes time after birth (postnatal testicular torsion-PsTT) 93% would perform an immediate investigation of the affected hemi-scrotum, 5% an urgent investigation, 1% a selective one, while 1% would not interfere surgically. 96% would perform a contralateral orchidopexy.

5% would use a scrotal incision and 25% the widely accepted by litterature inguinal incision. The reason for this approach is the treatment of a potentially different pathology such as testicular, paratesticular and metastatic malignancy.(39,40) With an inguinal incision pathology of processus vaginalis can be treated. Recent research demonstrates that most surgeons use a scrotal incision because they benefit from the pre-surgical ultrasound and it is technically easier. (41,42,43)

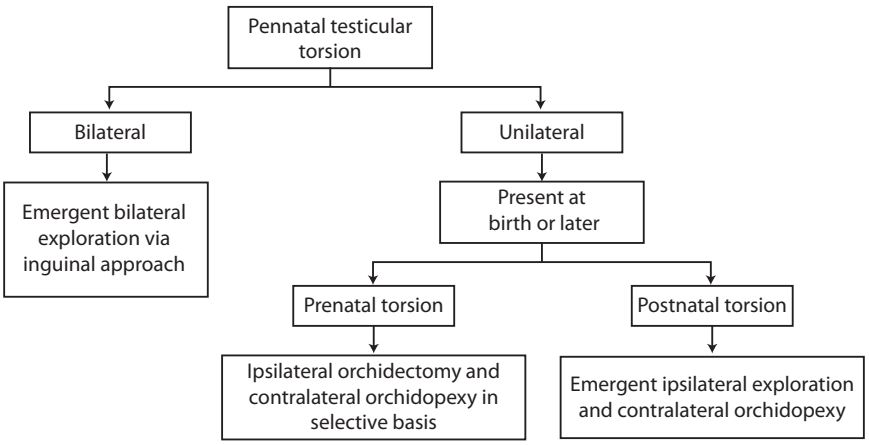
In their study, Rhodes HL, Corbett HJ et al (31) analysed 110 answers to the questionary sent to 148 pediatric urologists and pediatric surgeons, working as consultants in United Kingdom , about the management of PTT. 74,5% would investigate the pathologic hemiscrotum. If the testis was non-viable 71,9% would perform an orchidectomy and orchidopexy of the contralateral testis. 10% would investigate the pathologic hemiscrotum to prove the torsion without removing the necrotic testis or performing a contralateral orchidopexy. 8,1% would follow the steps above but with an orchidopexy. 1,8% would leave the non-viable testis at place without a contralateral orchidopexy. Finally, 17% would not interfere at all.

In the study of Kaye J, Levitt SB, et al (25) is demonstrated a very useful algorithm over the strategy of treatment of PTT (Table 2).

Table 1. *Classificasion of SFU and SPU members' answers.*

Disease	Immediate exploration (%)	Urgent exploration (%)	Elective operation (%)	No exploration %)	Use CDU for diagnostic confirmation (%)	Use inguinal incision (%)	Use scrotal incision (%)	Contra lateral orchiopexy (%)
PrTT	34	26	28	12	84	25	75	93
(b)PrTT	90	1	2	7	93	21	79	
PsTT	93	5	1	1	77	25	75	96

Table 2. Therapeutic strategy of PTT in according Kaye J, Levitt SB, et al.



In case of a non-viable testis we should take under consideration the possibility (>10%) of the development of malignancy in the future from the remaining germ cells (28,44). For bilateral PTT an orchidectomy is not recommended, with the hope that an endocrine activity of Leydig cells remains (45).

PROGNOSIS

The possibility of the survival of a tortued testis, either before birth or during the neonatal period, is 3-6%.(46) If we divide into two groups, PrTT and PsTT, then the percentage of tortued testicles remaining viable are importantly changing: in the group of PrTT it approaches 0% and in the group of PsTT it varies between 22-44%, especially when an immediate surgical intervention is attempted.(18,46,47)

Συστροφή όρχι κατά την εμβρυϊκή ζωή και τη νεογνική ηλικία (Περιγεννητική συστροφή όρχι)

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Με τον όρο περιγεννητική συστροφή όρχι (ΠΣΟ) αποδίδεται η εξωελυτροειδική –κατά κανόνα- συστροφή όρχι που συμβαίνει είτε κατά την εμβρυική ζωή (prenatal testicular torsion) είτε στη νεογνική ηλικία (postnatal testicular torsion). Αφορά το 10-22% των συστροφών όρχι που συμβαίνουν κατά την παιδική ηλικία. Τα κλινικά ευρήματα της ΠΣΟ όπως μπορούν να εκτιμηθούν μετά την γέννηση εξαρτώνται ουσιαστικά από τον χρόνο που συνέβη η συστροφή. Σε κάθε άρρεν νεογέννητο με διόγκωση οσχέου το οποίο έχει μελανόχρωμη εμφάνιση ενώ ο σύστοιχος όρχις έχει σκληρή σύσταση θα πρέπει να θεωρείται ως πιθανή η ΠΣΟ μέχρι απόδειξης του αντιθέτου. Σχετικά με την ενδεδειγμένη αντιμετώπιση της ΠΣΟ δεν υπάρχει ομοφωνία από τους συγγραφείς. Παραμένουν οι διαφωνίες αναφορικά με τον χρόνο και την μέθοδο αντιμετώπισης καθώς και για την αναγκαιότητα της προληπτικής ορχεοπηξίας του ετερόπλευρου όρχι. Στην παρούσα μελέτη, έπειτα από την διεξοδική αναδίφηση της πρόσφατης και σχετικά περιορισμένης βιβλιογραφίας, γίνεται προσπάθεια για τεκμηριωμένη αποσαφήνιση των ζητουμένων που παραμένουν.

Λέξεις-Κλειδιά: Αγομελατίνη, Γονοτοξικότητα, Χρωματιδιακές ανταλλαγές, Δείκτης ρυθμού πολλαπλασιασμού, Μιτωτικός δείκτης

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