CASE REPORT

A case of bilateral prenatal testicular torsion: Ultrasonographic features, histopathological findings and management

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Abstract
Objectives: The aim of this study was to demonstrate the ultrasonographic features of prenatal bilateral torsion of the testis, and its histological correlation and management.
Patient: A newborn presented at delivery with both testes enlarged, swollen and tender. Prenatal ultrasound (US) showed enlarged, hyperechoic testes. Colour Doppler US examination was performed.
Results: US revealed both testes to be heterogeneous. Colour Doppler US did not reveal any flow signal. On inguinal exploration both testes appeared necrotic. Histology showed recognizable seminiferous tubules and Leydig cells.
Conclusion: We believe that both testes should be left in situ after bilateral detorsion even if their macroscopic appearance is necrotic.

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Introduction

Bilateral intrauterine torsion of the testes is a rare cause of early testicular loss [1–3]. To date, 21 documented cases have been reported in the international literature [2]. Prenatal torsion, probably occurring around the 34th week of gestation, is almost exclusively extravaginal and presents as a hard, swollen, but not tender testis [4–6]. Colour Doppler ultrasonography is the diagnostic method of choice in a newborn with painless congenital scrotal swelling and a palpable mass [6]. Scrotal ultrasonography shows characteristic features in diagnosing prenatal torsion of the testis. The management of bilateral prenatal torsion is still controversial. Prenatal torsion noted at birth has not been considered a surgical emergency by some authors since the testes are not believed to be viable [1,2,7]. We present here a newborn with the presumptive in utero diagnosis of bilateral spermatic cord torsion. Management is also discussed in relation to histological findings.

Case report

A full-term 4100 g male newborn was delivered by primary cesarean section of a 28-year-old diabetic woman. The APGAR score was 9 and 10 at 1 and 5 min, respectively. Routine prenatal ultrasonography (US) (Ecocolourdoppler AU580AS, probe convex 3.5 MHz) at 34.5 weeks of gestation had showed a bilateral fetal hydrocele and moderately enlarged and hyperechoic testes (Fig. 1). There were no systemic symptoms. At physical examination, the scrotal skin was discromic and the testes appeared as an enlarged firm mass, not tender on palpation.

Neonatal US examination (5.12 MHz Philips ATL HDI 3000) showed both testes to be enlarged (right 1.9 cc; left 1.5 cc) with heterogeneous testicular texture, a thickened tunica albuginea, small bilateral hydrocele and thickening of the scrotal wall (Fig. 2). The colour/power Doppler did not reveal any flow signal. As a consequence, a bilateral testicular torsion was diagnosed and the newborn referred to emergency surgical exploration.

Transinguinal exploration revealed a twisted, black, necrotic-appearing right testis. The left testicle had also undergone extravaginal torsion. The gonads were untwisted, biopsied and fixed within the scrotum. On light microscopy, an extensive hemorrhagic and coagulative necrosis of the left testis was found, with diffuse, high-grade chronic inflammation. Surviving seminiferous tubules occasionally could be seen, including gonocytes and immature Sertoli cells (Fig. 3a). There was less significant damage to the right testis, whose parenchymal lobules showed some recognizable seminiferous tubules, although displaying focal regressive changes, and surviving endoluminal cells were present. Frequent intra- and extratubular calcium precipitates occurred. Leydig cell clusters were also evident inside the expanded and hemorrhagic interstitial stroma (Fig. 3b).

Figure 1 Prenatal US examination at 34th week of gestation: bilateral hydrocele (arrows); both testes were hyperechogenic and heterogeneous.

Figure 2 Neonatal US examination shows both testes to be enlarged (right 1.9 cc; left 1.5 cc). Note for both testes the heterogeneous pattern of parenchyma, a thickened tunica albuginea, small bilateral hydrocele, and thickening of the scrotal wall.
The postoperative course of the patient was uneventful. Fifteen days after surgery, scrotal US examination showed a reduction in the volume of both testes (right 0.67 cc; left 0.59 cc). In the right testis devolvement of a hyperechoic rim at the transitional zone next to the tunica albuginea was observed. Microcalcifications inside testicular parenchyma were also observed in both testes. On the left side a central liquefied area was also documented. The parents were informed of the high probability of sterility and possible need for exogenous androgen therapy to assure secondary sexual characteristics at puberty.

Discussion

Prenatal testicular twisting is an infrequent intrauterine event, with the unique presentation of an enlarged and firm scrotal testis, and is a cause of early testicular loss [2]. Most cases of intrauterine torsion have been reported to be unilateral [1,3,8], but bilateral lesions occasionally do occur [2,5,7].

The exact timing of prenatal testicular torsion is not known. Tripp and Homsy [5] reported a case of bilateral neonatal torsion suspected at the 34th week of gestation. Olguner et al. [7] and Hubbard et al. [9] reported a case of unilateral torsion at the 34th and 35th week of gestation, respectively. Prenatally detected hydrocele can be a sign of prenatal testicular torsion [5,8,9]. The precise etiology of intrauterine torsion of the testis is also unknown. A higher birth weight, trauma from a difficult labour or breech presentation, and an overactive cremasteric reflex have been implicated, although the most popular current theory is that the extreme mobility of the neonatal tunica vaginalis within the scrotum allows torsion to occur due to some violent cremasteric contraction occurring in utero or during delivery [1].

Gestational diabetes, causing a high neonatal weight, is a predisposing factor to prenatal testicular torsion [10]. In our case, the newborn was a full-term over the 90th percentile for weight.

Unlike postnatal torsion, newborns with prenatal torsion are generally asymptomatic and afebrile, and the only abnormality is an enlarged firm scrotal mass. Differential diagnosis in the neonate must include inflammatory and neoplastic processes affecting the testicle and the intrascrotal structures. Colour Doppler US is the diagnostic modality of choice in a newborn with a painless congenital scrotal swelling and a palpable mass [6]. Scrotal US shows characteristic features in diagnosing prenatal torsion of the testis [6]. These findings, in combination with absence of testicular flow on the affected side, are highly suggestive of prenatal torsion of the testis [6].

Many authors do not consider prenatal bilateral torsion, a surgical emergency, as examination of the removed testis some time after birth has revealed diffuse hemorrhagic parenchymal necrosis, hemosiderin and calcium deposition, and no viable germinative or Leydig cells [2,6]. However, we believe, on the basis of histological findings that emergency and conservative surgical treatment is effective. The reason for replacing a gangrenous organ is based upon the argument that the macroscopic appearance of the testis may not reflect its potential viability, especially of the endocrine (Leydig) cells which are known to be more resistant to ischemia [11–14]. The high proportion of fetal hemoglobin should also be taken into account as it partially prevents hypoxic/ischemic damage, but on the other hand can enhance both free iron release and peroxidative cell injury [15]. Moreover, the degree of ultrastructural modification caused by torsion depends mainly on the severity of the reduction in vascular supply rather than the length of duration of ischemia [16]. In fact, ischemic damage in bilateral
torsion is not always symmetrical. Finally, it is suggested that the survival of any tissue, however small, may have some cosmetic or psychological value, particularly in bilateral cases.

In conclusion, for bilateral prenatal testicular torsion, the preferred surgical approach is controversial [1,2]. We believe that both testes should be left in situ after bilateral detorsion via the inguinal approach, even if their macroscopic appearance is necrotic, in order to utilize the androgenic effects of surviving Leydig cells for as long as possible.

References