Ruptured Cystic Teratoma of the Testis in a Neonate

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Abstract: Neonatal testicular tumors are extremely rare. We report the case of a full-term male newborn with right hemiscrotal swelling found immediately after birth. The right hemiscrotal skin gradually turned dusky dark and then adhered to the hard scrotal contents. Missed right testicular torsion was suspected and emergency exploration was not attempted. Right hemiscrotal swelling became aggravated, so right orchiectomy and left orchidopexy were performed. During surgery, severe inflammation of the right testis and the right hemiscrotum were observed and diffuse hemorrhage with focal hematomas was noted within the removed right testis. Histopathology of the specimen revealed a ruptured mature cystic teratoma of the testis. The alpha-fetoprotein concentration was $729 \times 10^4 \mu g/L$ 10 days after the operation, and dropped to $185 \times 10^4$, $25 \times 10^4$, and less than $20 \times 10^4 \mu g/L$ 2, 5, and 8 months later, respectively. Postoperative abdominal and pelvic computerized tomography scans revealed neither lymphadenopathy nor distant metastasis. During a follow-up of 8 months, no evidence of tumor recurrence was found. Although extremely rare, testicular tumors should be included in the differential diagnosis of an enlarged hard or firm scrotal mass in the male neonate.

Case Report

A full-term male newborn delivered by a gravida 2 para 2 mother via normal spontaneous delivery with vacuum support was found to have right hemiscrotal swelling immediately after birth. Initially, no scrotal skin change suggestive of inflammation was noted and the scrotal contents were of usual consistency by palpation. Hydrocele was suspected and no treatment was given. However, within 24 hours, the right hemiscrotal skin turned dusky dark and the scrotal contents became hard and adhered firmly to the scrotal skin.

Scrotal sonography revealed an enlarged right testicle with heterogeneous echogenicity. Doppler ultrasonography showed equivocal right testicular blood flow. Missed right testicular torsion was then suspected. Because the opportunity for salvage of testicular torsion had passed, and because of the high complication rates for neonatal surgery, emergency exploration was not attempted and delayed exploration and contralateral orchidopexy was planned for 3 months later. The baby was discharged from the hospital 14 days after birth.

Right hemiscrotal swelling became aggravated after discharge, so he was readmitted for right orchiectomy and left orchidopexy 3 days after discharge. During the operation, severe inflammation of the right testis and the right hemiscrotum was observed. When the right testis was opened after removal, diffuse hemorrhage with focal hematomas was found. The postoperative course was uneventful except for prolonged wound healing of 15 days.

Histopathology of the specimen revealed a ruptured mature cystic teratoma of the testis (Fig. 1). Hair shafts with keratin and foreign-body reactions in an inflammatory background were demonstrated (Fig. 2). The concentration of alpha-fetoprotein was $729 \times 10^4 \mu g/L$ 10 days after operation, which dropped to $185 \times 10^4$, $25 \times 10^4$, and less than $20 \times 10^4 \mu g/L$.

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Postoperative abdominal and pelvic computerized tomography scans revealed neither lymphadenopathy nor distant metastasis of the testicular tumor. No evidence of tumor recurrence was found during 8 months of follow-up.

Discussion

The differential diagnosis of an enlarged hard or firm scrotal mass in the male neonate includes testicular torsion, inguinal hernia, hematoma, testicular tumor, meconium peritonitis, and idiopathic infarction, with testicular torsion being the most common [3, 7–9]. Testicular tumors are extremely rare in newborns at or before 1 month old [5–7]. In one study based on registration in the Prepubertal Testis Tumor Registry of the American Academy of Pediatrics, 22 neonatal testicular tumors were reported in a 12-year period [7]. Among these 22 tumors, there were six yolk sac tumors, six gonadal stroma tumors, six juvenile granulosa cell tumors, two gonadoblastomas, one hamartoma, and one teratoma.

Teratomas characteristically contain elements derived from all three embryonic germ layers and are composed of tissue elements foreign to the organ or anatomical site of origin [10]. In childhood, these tumors are notable for their diversity in anatomic location and biologic behavior [10]. They almost always arise in the sacrococcygeal, ovary, head and neck, retroperitoneum, mediastinum, testes, central nervous system, liver, abdominal wall, or back [10]. Various degrees of histologic differentiation are present in teratomas, and patients with mature teratomas, which are composed of well-differentiated tissue elements, always have a good prognosis. Testicular teratomas in children are most commonly found outside the newborn period and patients generally have a good prognosis even if solely treated using radical inguinal orchiectomy [7, 10]. However, neonatal testicular teratoma is so rare that no conclusion can be made regarding its pathologic characteristics and clinical outcome.

Neonatal testicular torsion can occur either in the prenatal or postnatal periods. In the newborn, testicular tumors are difficult to differentiate from prenatal testicular torsion since both represent themselves as enlarged, nontender, and firm or hard masses [2, 7, 8, 10]. The diagnosis of postnatal testicular torsion usually depends on physical examination, close observation, and strong clinical suspicion. Slow progression of testicular enlargement without apparent scrotal inflammation favors the diagnosis of testicular tumor. While prenatal testicular torsion may be asymptomatic, rapid increase in testicular size and conspicuous hemiscrotal reaction usually indicate postnatal testicular torsion [8, 11]. Severe scrotal inflammation associated with a neonatal testicular tumor, as manifested in the current case, is very unusual and might be attributed to testicular tumor rupture that possibly resulted from either a difficult delivery or spontaneous bleeding of the tumor. Recently, some investigators have advocated the use of scrotal Doppler ultrasound as a diagnostic modality for neonatal testicular torsion and their work shows promising results [12, 13].

In a newborn, when testicular torsion is suspected, emergency exploratory surgery should be performed within 4 to 6 hours [8, 9]. Generally, the gonad salvage rate in cases of neonatal testicular torsion is low, since most torsion develops prenatally and postnatal neonat-
tal testicular torsion is usually difficult to discover and correct within 6 hours of its development [9]. The optimal treatment of neonatal testicular torsion lasting longer than 6 hours remains controversial [8, 9, 11, 13]. Some authors suggest a delayed exploration or even no surgery, with close observation of the contralateral gonad to avoid surgical complications of neonatal surgery. Others advocate immediate orchietomy and contralateral orchidopexy to prevent the possibility of bilateral torsion, the negligence or delay in detecting other significant intrascrotal lesions such as neoplasms, and the detrimental effects of the torsed testis.

The present case suggests testicular tumors should be included in the differential diagnosis of neonatal testicular masses, even when clinical symptoms and signs favor testicular torsion.

References


