Prenatal diagnosis and successful postnatal treatment of huge sacrococcygeal immature teratoma: A case report with literature review

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Sacrococcygeal teratoma (SCT) is the most common congenital tumor of the newborn. Antenatal diagnosis of SCT is usually possible by ultrasound during the second or even earlier in the first trimester. It requires close fetal surveillance due to associated significant prenatal mortality such as fetal hydrops and high-output heart failure. Complete excision of the tumor is the mainstay of treatment and chemotherapy and radiotherapy can be conducted as adjuvant in malignant cases. Long-term follow-up is essential for assessment of recurrence and functional problems. We report a case of huge sacrococcygeal immature teratoma diagnosed by prenatal ultrasound, which successfully underwent postnatal complete excision of tumor after cesarean section. Clinical considerations are discussed.

Key words: Sacrococcygeal teratoma; Immature teratoma; Postnatal surgery

Introduction

Sacrococcygeal teratoma (SCT) is a rare subentity of germ cell tumor, but is the most common congenital tumor of the newborn, with an incidence of 1 in 35,000-40,000 live births and a female predominance in a 3:1 ratio. An excellent prognosis is followed if adequate surgical excision is prompt and complete. In contrast to newborn SCT, fetus with SCT remains at a high risk of perinatal complications and death. It requires close in utero surveillance due to their unpredictable growth and physiologic effects. Most SCTs are benign, but about 20% are malignant, though it is rare in the neonatal period (i.e. <1 month old). We report a case of huge sacrococcygeal immature teratoma diagnosed by prenatal ultrasound, which successfully underwent postnatal complete excision of tumor after cesarean section.

Case Report

A 33-year-old pregnant woman of 24 weeks’ gestation (parity: 1-0-0-1) visited the Samsung Medical Center for fetal anomaly counseling. She had no abnormal obstetrical and medical history. She was naturally conceived. The result of serum screening test (Triple marker test at 16 weeks’ gestation) was positive for Down syndrome (1:154) at another hospital, so amniocentesis was performed. Amniotic fluid analysis was reported to be normal (alpha-fetoprotein [AFP], acetylcholinesterase, chromosomal analysis). During the screening ultrasound, fetal sacrococcygeal teratoma was suspected and she was referred to our hospital. At our hospital (Fig. 1), about 7.0×6.6 cm sized mass with cystic and solid portion was observed at sacrococcygeal area by ultrasound. Polyhydramnios (amniotic fluid index, AFI: 26.03) was also observed. No other structural abnormality was identified. Doppler study for umbilical artery and middle cerebral artery was normal and hydropic features were not observed.
Fetal MRI was conducted additionally with a consultation to a pediatric surgeon for postnatal surgery. On fetal magnetic resonance imaging (MRI) (Fig. 2), 8.8×8.2 cm sized solid and cystic mass was observed in sacrococcygeal area and sacral bone defect was not observed with intact spinal canal. Urinary tract, brain, lung, liver and gastrointestinal tract had no abnormal finding. Routine prenatal check-up was conducted until 31^{\text{th}} week with no change of mass size and AFI and no evidences of heart failure on ultrasound. At 32^{\text{nd}} weeks, she was hospitalized due to preterm labor and betamethasone was administered for lung maturation. Uncontrolled preterm labor (unresponsive to tocolytics) resulted in emergent low segment transverse cesarean section. The baby was 2.43 kg male, Apgar score was 4 (1 minute) and 7 (5 minute).

Grossly, 12.5×9.0×8.5 cm sized tumor was located right below the perforated anus. The tumor was categorized as Altman’s type I according to its location. Meconium passed well. For further evaluation and postnatal management, baby was admitted to the neonatal intensive care unit. Brain/abdomen ultrasound, 2D echocardiography were conducted and associated anomalies were not found. Even though tumor showed presacral extension in postnatal ultrasound, it was not extended towards the spinal canal which was intact inside. The location of conus medullaris was normal as well. Tumor contained considerable part of solid component and it showed high vascularity on color Doppler.

Complete mass excision was successfully performed at the 4^{\text{th}} day after delivery (Fig. 3). Tumor weighs 700 g and on pathology report, it was consistent with sacrococcygeal immature teratoma with negative resection margin. Biopsy of coccygeal bone was negative for malignancy. On the chest CT and bone scan after the surgery, no metastasis was identified. Adjuvant therapy was not conducted. Full per oral feeding was possible on the 7^{\text{th}} day of birth. Because weight gain was steady and there were no postoperative complications, the baby was discharged on the 15^{\text{th}} day of birth. From then on, every 3 months of the first year and then every 6 months thereafter, ultrasound and AFP/β-hCG were serially followed and had no evidence of recurrence. After the birth, AFP/β-hCG marked 174 ng/mL/109 mIU/mL and marked 48,000 ng/mL/2 mIU/mL after surgery. It was all normalized on the follow-up. Up to recently (May 2010; 2 year follow-up), no postoperative complication and recurrence of tumor are observed. Functional problems such as feeding, defecation, urination, lower extremity weakness are also not observed and his current general condition is very tolerable.

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**Fig. 1.** The prenatal 2D/3D ultrasound images of tumor at Samsung Medical Center (24 weeks’ gestation).

**Fig. 2.** The images from fetal magnetic resonance imaging.