Diagnosis of sacrococcygeal teratoma using two and three-dimensional ultrasonography: two cases reported and a literature review

Mihaela Grigore1,2, George Iliev3

1Department of Obstetrics and Gynecology, University of Medicine and Pharmacy “Grigore T. Popa” Iasi, 2Medis Medical Centre, Iasi, 3Cuza Voda Hospital, Iasi, Romania

Abstract
The occurrence of a fetal tumor is rare, 50% of all fetal tumors being sacrococcygeal teratomas. Prenatal diagnosis of this condition is important for the monitoring of the affected fetuses and for establishing the mode and time of delivery. We describe two cases of fetal sacrococcygeal teratoma diagnosed by 2D- and 3D-ultrasound. Three-dimensional ultrasound proved to be useful both in the diagnosis and patient counseling. The combination of 2D- and 3D-ultrasound enables the diagnosis to be made during the first trimester of pregnancy.

Keywords: sacrococcygeal teratoma, 2D-ultrasound, 3D-ultrasound

Introduction
The occurrence of a fetal tumor is rare, and in half the cases, the tumors are sacrococcygeal teratomas (SCTs). The estimated incidence of SCT varies in the literature between 1 in 27,000 to 1 in 40,000 live births [1,2].

Before the routine use of ultrasonography in obstetrics, most fetuses with SCT were diagnosed at delivery. Advances in ultrasound have enabled accurate prenatal diagnosis of SCT, and in some cases, diagnosis can be made during the first trimester of pregnancy [2,3]. Ultrasound is useful not only for diagnosis, but also for monitoring the evolution of the tumor, detecting complications, and establishing the management. Three-dimensional (3D) ultrasound can be beneficial for monitoring the evolution of the tumor and for conveying to the parents a better understanding of this anomaly. We present two cases of SCT diagnosed antenatal with the aid of two-dimensional (2D)- and 3D-ultrasound.

Case 1
A 34-year-old patient, gravida 3 para 3, with 20 weeks of amenorrhea has presented for routine fetal examination. The 2D ultrasound revealed a bulky mass (97 x 68 x 67 mm) at the sacral area that was predominantly solid and had a few cystic components. The patient had a sonography in the first trimester of pregnancy, but at that time, the anomaly was not detected. No other fetal abnormalities were observed. A 3D ultrasound was performed with the volumetric transducer (Voluson 730 Pro, General Electric, Zipf, Austria). The 3D ultrasound depicted the relationship between the mass and the fetal pelvis and enabled the parents to better understand this anomaly (fig 1). A diagnosis of a SCT type I was formulated. Because of the tumor’s large size and the predominantly solid component, a poor prognosis was determined. The parents opted for a therapeutic abortion, and the diagnosis was confirmed thereafter.

Case 2
A 32-year-old patient, gravida 1 para 1, with 33 weeks and 4 days of amenorrhea was referred for an ul-
ultrasound from an ob-gyn specialist who had discovered at ultrasound a mass in the sacral area. A 2D ultrasound examination revealed a large mass of 44 x 44 x 41 mm starting from the sacral area. Using a volumetric transducer, 3D ultrasound was performed with Voluson 730 Pro (General Electric, Zipf, Austria) (fig 2). No other fetal abnormalities were detected. The patient returned two weeks later at 35 weeks and 2 days gestational age. The previously identified mass had slightly increased in size to 57 x 46 x 44 mm. After a consulting examination with the neonatologist, pediatric surgeon, and anesthesiologist, delivery was scheduled to be performed in a tertiary center. A cesarean section was performed at 38 weeks amenorrhea, and a newborn (3100 g) was delivered. The prenatal diagnosis of SCT type II was confirmed, and surgery was performed. The child had a normal development. Three years later, the women become pregnant again and delivered a healthy newborn (38 weeks, 3200 g).

Discussion

SCT arises from the pluripotent cell lines in Hensen’s node, which is located on the anterior surface of the sacrum or coccyx [4]. According to the Altman Classification of the Surgical Section of the American Academy of Pediatrics, SCTs are classified into four types [5]:

Type I- tumor is predominantly external projecting from the sacrococcygeal region and presenting with distortion of the buttocks.
Type II- tumor is predominantly external, but has a large intrapelvic component.

Type III- tumor is predominantly intrapelvic with a small external buttock mass.

Type IV- tumor is entirely internal with no external component.

All four types may have an intraspinal component with neurologic impairment. The SCT can be cystic, solid, or mixed. Predominantly cystic lesions are usually benign [6]. In our cases, the first one was type I predominantly solid, or mixed. Predominantly cystic lesions are usually benign and carry a good prognosis. However, the type of tumor significantly influences the prognosis. For example, type III and type IV tumors are more likely to be malignant, while type I and type II are more likely to be benign [6].

In the majority of cases (80%) the tumor is benign, but in up to 20% of cases, SCT can be malignant [7]. Although most cases are benign, SCTs are associated with high morbidity and mortality rates because of preterm delivery and complications, such as malignant invasion, hemorrhage into the tumor, obstruction of umbilical flow, high output cardiac failure, hydrops fetalis, and bladder outlet obstruction [8,9]. Because some of these complications can be detected prenatally and treated appropriately, prenatal diagnosis of SCT is very important.

Advances in ultrasound have enabled accurate early diagnosis of SCT, even in the first trimester of pregnancy. Sonography can establish the type of the tumor according to Altman classification and can also determine whether SCTs are cystic, solid, or mixed. Ultrasound scans are also necessary to monitor the tumor size, extension into adjacent structures, tumor vascularity, and evidence of cardiac failure. Poor prognosis indicators are predominantly solid components, hydrops, and diagnosis before 30 weeks’ gestation [9-11]. Serial fetal ultrasounds are indicated whenever a sacral mass is discovered. Ultrasonography is also useful in determining time and mode of delivery [11]. In our cases, the diagnosis was established at 20 weeks for the first case and 33 weeks for the second one.

Doppler ultrasound is useful for detecting reverse blood flow in the umbilical arteries indicating arteriovenous shunting through the tumor. According to Olu-toye, abnormal umbilical artery waveforms are an indication for utero intervention or inducing delivery to prevent fetal demise [12].

Recently, several papers have been published regarding the role of 3D ultrasound in the diagnosis of SCT. Three-dimensional ultrasound has been used to monitor volume changes of the tumor, assess tumor extension by visualizing the skeleton and pelvic bones, and mapping the blood supply [7,13,14].

Using 3D ultrasound, the diagnosis of SCT has been made possible even during the first trimester of pregnancy [2,3]. Also, 3D ultrasonography has been particularly useful in counseling the parents and describing the lesion to them. In our cases, 3D ultrasound with render mode helped us to evaluate the morphology of the tumor and the relationship with the pelvis and skeleton.

Magnetic resonance imaging (MRI) has also been described as an adjunct method for diagnosing SCT. MRI could depict with great accuracy the intrapelvic extension of the tumor, but until now, its use has been reported only during the third trimester of pregnancy [15,16].

The diagnosis of a SCT needs a multidisciplinary approach to establish the prognosis and optimize the outcome. The perinatal management of SCT requires communication between obstetricians, neonatologists, and pediatric surgeons [8].

**Conclusion**

SCT can be diagnosed during pregnancy. In some cases, the diagnosis is possible during the first trimester of pregnancy. Because of this possibility, screening for chromosomal anomalies conducted at 11-13 weeks of amenorrhea should be used as an opportunity to detect SCT as early as possible.

**References**