Ultrasonographic Evaluation Of Fetal Sacrococcygeal Teratoma: Mapping Out The Vascularity

Fetal Sakrokoksigeal Teratomda Ultrasonografik Değerlendirme: Vasküleritenin Detaylandırılması

Mehmet Özgür AKKURT¹, And YAVUZ¹, Mekin SEZİK¹, Gökhan KARAKOÇ²

- ¹ Süleyman Demirel University, School of Medicine, Department of Perinatology, Department of Obstetrics and Gynecology, Isparta, Türkiye
- ² Maltepe Hospital Department of Obstetrics and Gynecology, İstanbul, Türkiye

ABSTRACT

Sacrococcygeal teratoma (SCT) is one of the most common fetal neoplasms with an incidence of around 1/27000 live births with female-to-male ratio of 4:1. A 37-year-old multigravida was referred for evaluation of a fetal presacral mass that was seen on ultrasonography (US) at 21 weeks' gestation. A female fetus presenting with a heterogeneous, solid mass with cystic components, measuring approximately 8x7x7.5 cm starting from sacral region was detected. The couple elected to terminate the pregnancy, and postnatal findings confirmed the diagnosis of SCT. Three dimensional ultrasound is a valuable adjuvant to routine B-mode sonography for antenatal diagnosis of SCTs and can define the degree of involvement of the sacrum. It is also a useful tool to map out the vascularity of fetal SCTs and thus allows visualization of the blood flow between SCTs and fetal circulation and enables parents to visualize the lesion.

Keywords: Doppler ultrasonography, prenatal ultrasonographic diagnosis, teratomas, three-dimensional imaging

ÖZ

Sakrokoksigeal teratom en sık gözlenen fetal tümörlerdendir. İnsidansı 27.000 canlı doğumda bir olup, kızlarda yaklaşık 4 kat daha fazla görülür. 37 yaşında, multigravida, 21 haftalık gebeliği olan hasta dış merkezden presakral kitlenin araştırılması için perinatoloji kliniğimize yönlendirilmiştir. Kız fetuste sacral bölgededen başlayan yaklaşık 8x7x7.5 cm lik heterojen, yer yer kistik komponentler içeren solid bir kitle izlendi. Aile gebeliğin sonlandırılmasını seçti ve postnatal tanı SCT ile uyumluydu. 3 boyutlu görüntüleme, SCT tanısında ve sakrumun invazyonun değerlendirilmesinde rutin B-mod sonografiye yardımcı olur. Ayrıca tümörün vasküleritesinin detaylandırılmasında, fetal dolaşım ile tumor arasındaki kan akımının görüntülenmesinde ve ailenin lezyonu, gözünde daha iyi canlandırmasında kullanışlı bir araçtır.

Anahtar Kelimeler: Doppler ultrasonografi, prenatal ultrasonografik tanı, üç boyutlu görüntüleme, teratom

Introduction

Fetal tumors are very rare and may occur in any fetal system during antenatal period. Sacrococcygeal teratoma (SCT) is probably the most common fetal neoplasm. Incidence of this tumor isapproximately 1 in 27000 live births (1). SCTs are four times more common in females than in males (2). The embryologic origin of germinal neoplasms is not known exactly. SCTs are believed to arise from pluripotent cells originating from the Hensen's node. More than 50% of SCTs contain calcification and ossification. They present as cystic, solid or mixed masses. Most SCTs are benign; however, up to 38% can contain malignant components especially in SCTs with entirely pelvic component (3).

Antenatal diagnosis of SCTs during routine midtrimester sonography is possible. This allows appropriate visualization of the tumor, its vascularity, and nature (i.e. cystic or solid) with the help of novel techniques such as 3-dimensional (3D) ultrasound including rendering and tomographic ultrasound imaging (TUI). Since these tumors are associated with high pre- and postnatal morbidity and mortality, 3D sonography techniques can enable the obstet-

rician and the parents to visualize the lesion and figure out the pathological alteration more clearly. Here, we report a case of SCT diagnosed at 21 weeks of gestation with emphasis on Doppler vascular mapping and 3D ultrasound visualization.

Case Report

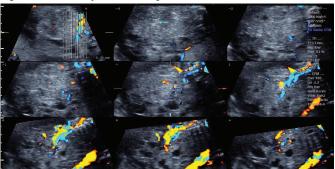
A 37-year-old multigravida was referred for evaluation of a fetal presacral mass that was seen on ultrasonography (US) at 21 weeks' gestation. A female fetus presenting with a heterogeneous, solid mass with cystic components, measuring approximately 8x7x7.5 cm starting from sacral region was detected. Prenatal SCT was diagnosed by B-mode and color Doppler two-dimensional US (Figure 1). Furthermore, three-dimensional (3D) Doppler US was used to map out vascularity of the tumor. Ultrasound volumes were acquired and were transferred to external imaging software (4D View, GE Healthcare, United States) for further offline analysis. "Glass body" and TUI images are shown in Figure 2. After reviewing the images and consultation, the couple elected to terminate the pregnancy. Postnatal findings confirmed the diagnosis of SCT,

Yazışma Adresi/ Correspondence Address: And Yavuz Süleyman Demirel Üniveristesi Tıp Fakültesi 32260 Çünür, İsparta, Türkiye Tel/Phone: +90 246 211 92 39 E-mail: andyavuz@gmail.com Geliş Tarihi/ Received: 19/01/2015 Kabul Tarihi/ Accepted: 04/02/2015 although the couple did not opt for postmortem examination.

Figure 1: B-mode and color Doppler two-dimensional US view



Figure 2: "Glass body" and TUI images



Discussion

Prenatal SCT can be diagnosed by 2D and 3D ultrasound with increasing use of prenatal sonography. However, a study reported an antenatal diagnosis rate of 44% percent (4). Prenatal diagnosismay be crucial, since sonographic findings can predict the prognosis, especially in cases with a dismal outcome. Hence, antenatal sonographic findings may provide important prognostic information (4,5). Generally poor prognosis such as fetal hydrops, perinatal death, need for fetal intervention were reported to be associated with the following antenatal findings: Large lesions (>10 cm), a tumor volume to fetal weight ratio (TFR) of >0.12 on ultrasound before 24 weeks and a TFR of \geq 0.11 before 32 weeksof gestation, solid tumors, polyhydramnios, and placental thickening (4,6).

In cases of fetal SCT, congestive heart failure and hydrops can develop prenatally. Postnatally these neonates have 9% risk of mortality in surgery and are at risk for long term neurological impairment of both sensory and motor function (7). Therefore, appropriate fetal diagnosis and counseling are essential steps for the obstetrician and the parents to decide on the course of pregnancy.

3D ultrasound is a valuable adjuvant to routine 2D sonography for antenatal diagnosis of SCTs and can define the degree of involvement of the sacrum. It is also a useful tool to map out the vascularity of fetal SCTs and thus allows visualization of the blood flow between SCTs and fetal circulation and enables parents to visualize the lesion (8). We suggest that 3D ultrasound including surface rendering and Doppler acquisition are necessary steps for the differential diagnosis and counseling of prenatal SCT cases. For example, surfa-

ce rendering allows parents to conceptualize the lesion in a realistic manner. Moreover, "glass body" visualization that includes Doppler interposed on 3D surface rendering helps to map out the lesion with proper details, serving as a precise tool for predicting the short- to intermediate-term outcome. The visualization of the vascularity may also aid in defining the suitability of the case for fetal therapy, i.e. fetoscopic embolization of the feeding vessel. However, fetal therapy is still experimental at this stage and is not widely available especially in the developing World.

In conclusion, 3D Doppler US is a valuable tool for the antenatal diagnosis and counseling of SCT. It can also be used to map out the vascularity of fetal SCTs and enables the physician and parents to visualize the lesion.

References

- Swamy R, Embleton N, Hale J. Sacrococcygeal teratoma over two decades: birth prevalence, prenatal diagnosis and clinical outcomes. Prenat Diagn 2008; 28:1048.
- Rescorla FJ, Sawin RS, Coran AG, et al. Long-term outcome for infants and children with sacrococcygeal teratoma: a report from the Childrens Cancer Group. J Pediatr Surg 1998; 33:171.
- Altman RP, Randolph JG, Lilly JR. Sacrococcygeal teratoma: American Academy of Pediatrics Surgical Section Survey-1973. J Pediatr Surg 1974; 9:389.
- Tailor J, Roy PG, Hitchcock R, et al. Long-term functional outcome of sacrococcygeal teratoma in a UK regional center (1993 to 2006). J Pediatr Hematol Oncol 2009; 31:183.
- Rodriguez MA, Cass DL, Lazar DA, et al. Tumor volume to fetal weight ratio as an early prognostic classification for fetal sacrococcygeal teratoma. J Pediatr Surg 2011; 46:1182.
- Shue E, Bolouri M, Jelin EB, et al. Tumor metrics and morphology predict poor prognosis in prenatally diagnosed sacrococcygeal teratoma: a 25year experience at a single institution. J Pediatr Surg 2013; 48:1225.
- 7. Wee WW, Tagore S, Tan JV, Yeo GS. Fetal sacrococcygeal teratoma: extremes in clinical presentation. Singapore Med J. 2011; 52:118-23.
- C.-P. Chen, J.-C. Shih Prenatal visualization of the vasculature of fetal sacrococcygeal teratoma by three-dimensional color power angiography Ultrasound in Obstetrics & Gynecology 2002; 20:636-637.