Acta Scientific Women's Health (ISSN: 2582-3205)

Volume 2 Issue 8 August 2020

Case Report

Fetal Sacrococcygeal Teratoma Requiring Emergency Hysterotomy in Second Trimester

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DOI: 10.31080/ASWH.2020.02.0129

Received: May 14, 2020

Published: July 30, 2020

Abstract

Sacrococcygeal teratoma (SCT) is the commonest fetal tumor. We report a case of fetal SCT requiring emergency hysterotomy at 21+4 weeks of gestation. A 22 years primigravida, with prior uneventful antenatal care, presented at 21+4 weeks of gestation with on and off pain abdomen and back for 3 days associated with passage of show for 1 day. Vital signs were stable and her abdomen was tense and tender with fundal height of 30 weeks. She was in early labor. Urgent USG revealed a live intrauterine fetus with features suggestive of huge fetal SCT for which she underwent emergency hysterotomy.

Keywords: Fetal; Hysterotomy; Labor; Sacrococcygeal Teratoma; Tumor

Introduction

Fetal SCT is the commonest congenital tumor with the incidence of 1/40,000 live births, with a male to female ratio of 1:3 [1]. It arises from the pluripotent cells of the caudal end (Hensen's node) of the primitive streak and consists of tissues arising from all three germ layers. Failure of degeneration of Hensen's node leads to tumor growth [2]. These are generally solid or complex with only 15% being entirely cystic [3]. Though, the majority of these tumors are histologically benign, they cause increased perinatal morbidity and mortality owing to prematurity, labor dystocia, fetal hydrops secondary to shunting of fetal blood to the tumor bulk or fetal exsanguination secondary to tumor rupture during delivery. Management depends on the age of gestation at diagnosis, presence of hydrops/polyhydramnios/placentomegaly, fetal lung maturity, availability of prenatal and neonatal surgical expertise and facilities. Termination of pregnancy when opted before fetal viability is usually done medically. Here, we report a case of pregnancy with a huge fetal sacrococcygeal teratoma, presenting with acute abdomen that required emergency hysterotomy.

Case Report

A 22 years primigravida, married for 7 months, with prior routine uneventful antenatal care at a private clinic presented at 21^{+4} weeks of gestation according to her LMP (7^{th} March, 2018) with the complaints of abdominal and back pain on and off for 3 days; and blood mixed, mucoid PV discharge for 1 day. She had taken folic acid supplement (5 mg daily) from 6^{+5} weeks to 14 weeks of gestation, following which iron and calcium supplementation was started. Her antenatal investigations were normal and her USG at 7^{+5} weeks confirmed with a single intrauterine fetus of 8 weeks. Nausea and vomiting of pregnancy was managed with ranitidine, ondansetron and multivitamin for 2 weeks. Fundal height was 3 weeks larger than date with regular fetal heart sound at 19^{+3} weeks. However, she had not experienced quickening yet. She did not have any significant personal, past medical and surgical history except for emergency appendectomy 2 years back. There was no history of any chronic medical illness or birth of anomalous babies in the

family. She received 1 dose of Inj. DT at 19^{+3} weeks when she was asked to follow up with fetal anomaly scan.

At presentation in ER at 21^{+4} weeks, she was in pain. Her vital signs were normal. Abdomen was tense, tonically contracted and mildly tender with the fundal height of 30 weeks size. Fetal heart sound couldn't be localized with doppler. On vaginal examination, she was found to be in early labor.

With the suspicion of placental abruption with fetal demise, blood investigations and ultrasonography (USG) were done. Routine hematological and biochemical investigations were normal with Hb of 13.9 gm/dl. USG revealed single live intrauterine fetus (EFW 600 grams) in cephalic presentation, corresponding to 22^{+3} weeks POG with a large heteroechoic mass measuring 14.7 x 11 x 10.4 cm^3 arising from the sacrococcygeal region with multiple foci of calcification and minimum vascularity within. Predominant portion of the mass was external to the pelvis, without intrapelvic extension. Placenta was anterior upper uterine without retro-placental clot and amniotic fluid volume was normal. Patient opted for termination of pregnancy and underwent emergency hysterotomy for preterm labor with huge fetal SCT.

An alive female fetus with low APGAR scores, weighing 1000 grams was delivered with fleshy growth of 15 x 11 x 10 cm arising from the fetal sacral region with bony and cartilaginous tissues within. Anal opening was absent. Placenta was anterior lower uterine with placentomegaly and weighed 500 grams. Liquor was abundant. Baby died immediately after birth. Patient's postoperative period was uneventful and she was discharged on 3rd postoperative day on oral antibiotics and analgesic. Histopathological examination (HPE) confirmed SCT.

Discussion

Majority of the cases of congenital SCT may be asymptomatic prenatally, diagnosed incidentally during routine anomaly scan. Fundal height larger for date may be the only suggestive finding, like in our case. Ultrasonography helps in diagnosis in the majority of cases. About 80% of diagnoses were made by second trimester fetal anomaly scan and the median age for detection was 20^{+3} weeks in a population based cohort study conducted by Ayed A., *et al*. over an 18-year period (1995 - 2012) in the west midlands re-

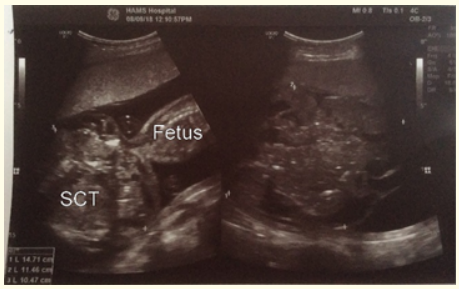

Figure 1: USG picture of heteroechoic mass arising from the sacrococcygeal region of the fetus with multiple foci of calcification.

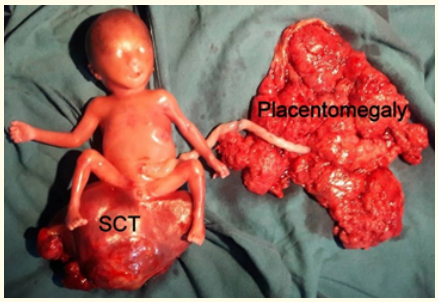

Figure 2: Ventral view of the fetus showing sacrococcygeal tumor, absent anal opening and large placenta.

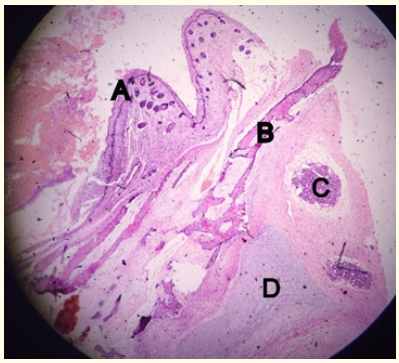

Figure 3: HPE showing tissues of different cell lines. A- Stratified squamous epithelium with hair follicles, B- Bone, C- Gland, D- Cartilage.

gions, UK [4]. Ultrasound diagnosis may be supported by findings from MRI or Doppler USG.

Prognosis of fetus with antenatally diagnosed SCT correlates inversely with tumor size, tumor growth rate, time of gestation at which the tumor is diagnosed and the presence of placentomegaly and/or hydrops fetalis [5]. The risk of preterm delivery correlates with the size of the tumor and the presence of hydramnios [6] features which could have triggered preterm labor in our case.

When the tumor is diagnosed at an early age of gestation, termination of pregnancy is usually offered, as the fetal prognosis is usually poor. Fetal surgical options include radiofrequency ablation of the vascular shunts to tumor or in-utero resection of the tumor if facilities are available. In the absence of placentomegaly and fetal hydrops, serial ultrasonographic follow up of the fetus should be done until fetal lung is mature, when elective cesarean section can be performed [7]. Cesarean delivery should be performed prior to onset of labor if the SCT is solid or > 5 cm in diameter [6]. Cesarean delivery in this case requires a large vertical uterine incision, so as to minimize traumatic delivery that may trigger massive haemorrhage [8].

In our case, the patient presented to emergency in early stage of labor. The presentation was early (21^{+4} weeks) i.e. before the age of fetal viability in our setup. Tumor was predominantly solid and the size was large, largest dimension being nearly 15 cm. Polyhydramnios and placentomegaly was evident intraoperatively. Though detailed evaluation of fetal anomaly couldn't be done, absence of anal opening could be a part of associated anomalies. All these findings supported our decision of termination of pregnancy. Augmentation of labor with oxytocin could have been an alternative to emergency hysterotomy. But, this could have led to labor dystocia owing to large solid tumor with multiple areas of calcification. So, we believe emergency hysterotomy through a low transverse uterine incision is justifiable in this case. Had we done USG at 12 - 13 weeks, SCT could have been detected early, which could have allowed for medical termination of pregnancy, avoiding the necessity of hysterotomy.

Conclusion

SCT is the most common fetal tumor. Its prognosis depends on numerous factors like: rate of growth of tumor, size of tumor, associated other anomalies, polyhydramnios, hydrops etc. At places like ours where there is neither availability nor affordability for fetal surgical services by the general public, prenatal detection can immensely help the parents in decision making. When fetal SCT is detected early, pregnancy can be terminated medically. Late detection may allow the SCT to grow to a large size, necessitating hysterotomy like in our case. This highlights the importance of early anomaly scan at 11 - 13 weeks of gestation, a procedure which is not yet widely practiced in our parts of the world.

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