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CASE REPORT

Case Report of a Rapidly Growing Sacrococcygeal Teratoma: A Planned Preterm Cesarean Delivery May Improve Fetal Outcome

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Abstract

Sacrococcygeal teratoma (SCT) is the most commonly diagnosed congenital tumor during the neonatal period. The outcome of SCT is dependent on the growth rate, the presence of solid components in cancer, and the presence or absence of high flow cardiac involvement of the fetus. This case report presents the management and outcome of a pregnant woman carrying a fetus with a rapidly growing SCT. Confirmation of diagnosis was based on the universal prenatal ultrasound examination standards. Eventually, the patient delivered a girl weighing 2.9 kg, including the neoplasm, by a planned Cesarean section. The newborn received one dose of surfactant and underwent complete resection of the teratoma on the second day with a favorable outcome. This case report aims to show that rapidly growing SCT requires frequent antenatal follow-up with serial prenatal ultrasound examinations, optimum timing, mode, and appropriate place of delivery for a better fetal outcome.

Keywords: Prenatal diagnosis, Preterm delivery, Rapidly growing neoplasm, Sacrococcygeal teratoma, Surgical resection of teratoma

Introduction

The prevalence of sacrococcygeal teratoma (SCT) in neonatal neoplasms is reported to be 1 in every 35,000 to 40,000 live births. In addition, approximately 80% of the affected infants are female.[1, 2, 3] Owing to the introduction of universal

prenatal ultrasound screening and improvement in ultrasound techniques, over half of SCTs, are diagnosed during the antenatal period.[4] Most of these tumors are benign with good outcomes.[5] Moreover, a rapidly growing teratoma requires either fetal therapy or premature delivery to avoid complications. Several case reports and peer-reviewed literature have presented unfavorable effects following fetal medicine performed for rapidly growing teratomas with fetal cardiac involvement.[6] This case report presents the management and outcome of a pregnancy with a rapidly increasing SCT delivered prematurely at 34 weeks and underwent surgical resection on the second day of life with good clinical and functional outcomes.

Case presentation

A 31-year-old woman (G2, P1+0) presented with no history of complications during previous pregnancy and delivery. The fetus was diagnosed incidentally with SCT at 20 weeks of gestation during her second pregnancy by a routine mid-trimester anomaly scan. The ultrasound examination described a mixed mass of 5.36 cm x 4.03 cm in the distal portion of the fetal spine, with no other apparent anomalies. Amniotic fluid index (AFI) and other findings were unremarkable. The mass was provisionally diagnosed as SCT and was verified by a second sonographer as a 4.6 cm exophytic mixed solid-cystic mass at the base of the spine with a minor component anterior to the sacrum. The diagnosis was suggestive of immature SCT, mild polyhydramnios, fetal size appropriate for dates, and no other structural abnormalities. The fetus was monitored through ultrasound scans and bi-weekly follow-up sessions.

At 23 weeks of gestation, the mass enlarged to 7.49 cm x 6.79 cm and increased amniotic fluid volume. A multidisciplinary team of obstetricians, pediatric surgeons, and neonatologists decided to deliver the patient at 37 weeks by cesarean section. At 32 weeks, the ultrasound revealed that the teratoma had further enlarged to 11.5 cm x 11 cm, with significant vascularity (Figure 1a and 1b). The AFI was found to be 31 cm indicating significant polyhydramnios. Due to the increasing size of the

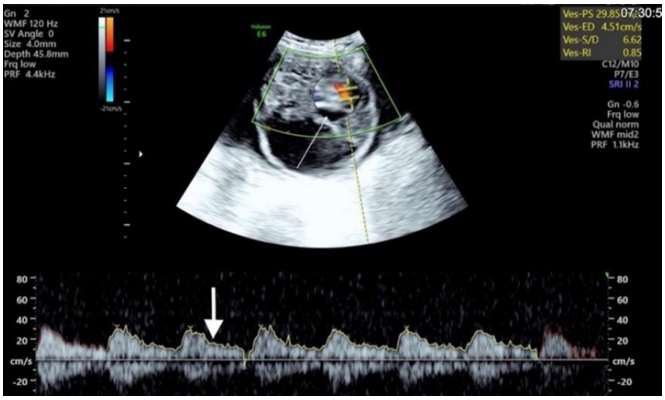

Figure 1a

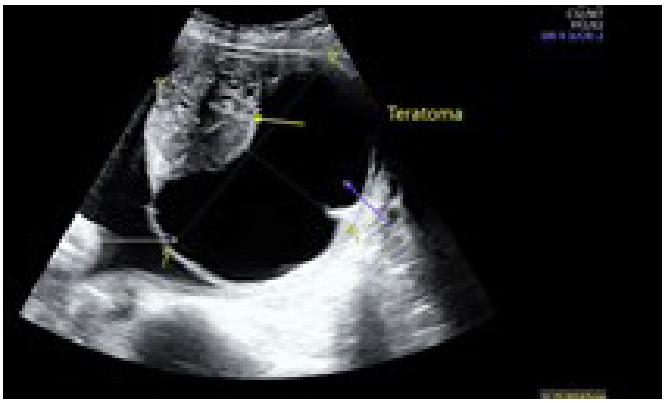

Figure 1b

Figure 1: US image of SCT at 32 weeks; a) Doppler flow inside the teratoma and b) solid and cystic components.

mass and polyhydramnios, cesarean delivery was preponed to 34 weeks gestation after antenatal corticosteroid. The decision was made to avoid further complications such as premature pre-labor rupture of membranes (P-PROM), preterm labor, cord prolapse, abruption of placenta, birth injury, intra-tumor hemorrhage, and tumor rupture, as well as to avoid emergency delivery.

The delivery outcome was an otherwise normal baby with a birth weight of 2.9 kg, including the neoplasm. The teratoma was found to be mixed with solid and cystic components along with bruise, ulceration, and necrosis over the tumor and occupying the whole sacral area (Figure 2). The mother was discharged on the second day of puerperium in stable condition. Immediately after delivery, the newborn's alpha-fetoprotein (AFP) exceeded 1000 ng/ml, and free beta HCG was 2 ng/ml. The newborn received one dose of surfactant for respiratory distress syndrome. An ultrasound of the abdomen was done to see the extent of the SCT.

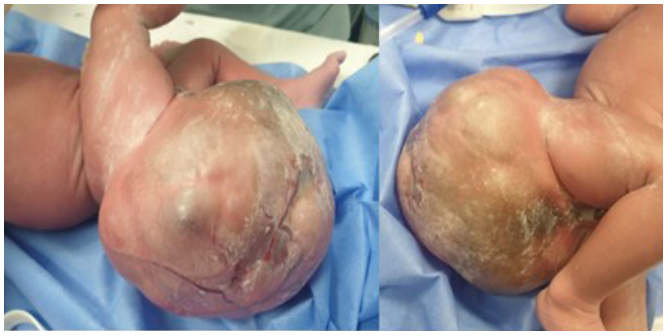

Figure 2: SCT immediately after delivery

It was found to be type 1 (Primarily external or has a minimal presacral component) according to the AAPSS classification.

Surgical removal of SCT was performed after 48 hours of delivery with an uneventful intraoperative and postoperative period. After resection of the teratoma, fetal weight was 2.2 kg. The resected mass was 10 cm x 9 cm x 4.5 cm consisting of solid, cystic, and hemorrhagic areas. The histopathology revealed a grade 3 immature teratoma with no malignant somatic components. Post-operatively, the baby received one unit of blood transfusion. Post-operative plain x-ray and ultrasound of the abdomen showed unremarkable findings. Initially, she needed anal dilation following surgery to enhance defecation. The baby was placed under observation with serial AFP and ultrasound examinations.

On follow-up at six months of age, the baby was found to have regular bowel and bladder functions. The operation site appeared to have a nearly typical configuration with a well-healed and cosmetically acceptable scar (Figure 3a and 3b). AFP dropped to 14 ng/ml, and an abdomen and renal system ultrasound revealed normally growing structures.

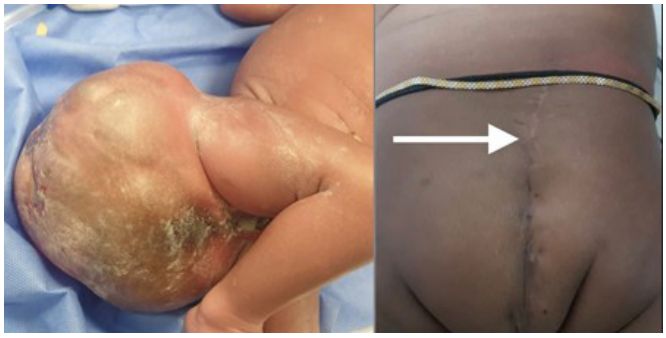

Figure 3: Comparison of the lesion site; a) before operation and b) 5 months after the operation, arrow indicates the scar.

Discussion

Most SCT cases are diagnosed between 22 to 33 weeks by an ultrasound examination during routine antenatal screening. The sacrococcygeal region is the most common site, and the average diameter of an SCT is 8 cm.[2] Studies were conducted to predict that prognostic factors of antenatally diagnosed SCT are mostly inconsistent.[7] Factors such as the size of the tumor, the presence of vascularity, and solid components in the tumor were described as most important.[8,9] Nevertheless, the relationship between the tumor size and prognosis has not been consistently supported by the authors.[9,10] The reported mortality due to SCT in the literature varies from 25% to 50%.[7,10] Long-term outcome in a fetus with SCT is excellent if diagnosed postnatally, whereas mortality is high when diagnosed during pregnancy.[8,11,12] In the presented case, SCT was diagnosed at 20 weeks of pregnancy and was rapidly growing, reaching 11 cm at 34 weeks, which made it considerably difficult to approximate the estimation of the actual size at term. The woman was kept under frequent follow-up as the teratoma had mixed solid and cystic components and high vascularity on sonographic examination to identify the risk of decompensation. [9, 10, 13, 14] Among the treatment modalities, either preterm delivery or fetal intervention like open fetal surgery to remove the tumors, radiofrequency ablation, major vessel laser ablation, vessel alcohol sclerosis, or amnioreduction are performed to prevent fetal complications and death.[12,15] Fetal interventions are often considered only when a highly trained surgeon skilled in these procedures is available and fetal compromise is detected before the age of viability; however, complications like intrauterine death and preterm birth may still occur.[15] On the other hand, early delivery, which may even be a preterm delivery followed by postnatal surgery, is considered one of the best options to prevent intrauterine death and avoid complications related to fetal surgery.[15]

As there were unfavorable prognostic factors like rapid growth, presence of vascularity, and solid components of the teratoma, in this case, the fetus was delivered by planned cesarean section at 34 weeks after arranging an elective postnatal surgery. Although vaginal delivery is not contraindicated,

cesarean section is strongly recommended for teratoma more than 5 cm in diameter to avoid dystocia, birth injury, intratumor hemorrhage, and tumor rupture.[2] Prognosis of a child with SCT depends on neoplasm recurrence, which is more common when the SCT is removed incompletely and the presence of malignant change in the neoplasm.[16] The baby's follow-up with ultrasound and AFP represented the adequate removal of SCT. The infant of the presented case had an excellent prognosis. The risk of recurrence is reduced because histopathology did not find any malignant changes in the neoplasm, and the teratoma was removed entirely.

Conclusion

This case report emphasizes the importance of ultrasound scans for early detection and follow-up of SCT through serial ultrasonographic examinations. In addition, it highlights the effectiveness of a multidisciplinary approach to offer the best neonatal outcomes by deciding on optimal timing, mode, and place of delivery. Even though prematurity is a concern, a fetus with SCT can be delivered a few weeks earlier than term and undergoes teratoma resection shortly after delivery. Therefore, it is crucial to balance preterm birth complications and the risk of intrauterine fetal demise with the continuation of pregnancy.

Conflict of interests

None to declare

Ethical approval

The case report was approved by the Head of Scientific Research and Development Department at King Hamad University Hospital, Bahrain.

Informed Consent

Informed consent was provided from the patients to use information and photography.

Author Contributions

All authors share equal efforts towards (1) Substantial contribution to conception and design, acquisition, analysis, and interpretation of data; (2) Drafting the article and revising it critically for important intellectual content; and (3) Final approval of the manuscript version to be published.

Data Availability

The authors declare that data supporting the findings of this case report are available within the article.

References

1. Tuladhar R, Patole SK, Whitehall JS. Sacrococcygeal teratoma in the perinatal period. PMJ. 2000; 76:754-759. https://doi.org/10.1136/pmj.76.902.754
2. Hu Q, Yan Y, Liao H, et al. Sacrococcygeal teratoma in one twin: a case report and literature review. BMC Pregnancy Childbirth. 2020; 20: 751. https://doi.org/10.1186/s12884-020-03454-1
3. Adekola H, Mody S. Bronshtein E, et al. The clinical relevance of fetal MRI in the diagnosis of Type IV cystic sacrococcygeal teratoma–a review. Fetal Pediatr Pathol. 2015; 34(1): 31–43. https://doi.org/10.3109/15513815.2014.949934
4. Swamy R, Embleton N, Hale J. Sacrococcygeal teratoma over two decades: birth prevalence, prenatal diagnosis, and clinical outcomes. Prenat Diagn. 2008; 28: 1048–1051. https://doi.org/10.1002/pd.2122
5. Kum CK, Wong YC, Prabhakaran K. Management of fetal sacrococcygeal teratoma. Ann Acad Med Singap. 1993; 22(3): 377-80. PMID: 8373123.
6. Lee MY, Won HS, Hyun MK, et al. Perinatal outcome of sacrococcygeal teratoma. Prenat Diagn. 2011; 31(13):1217-21. https://doi.org/10.1002/pd.2865
7. Flake AW. Fetal sacrococcygeal teratoma. Semin Pediatr Surg. 1993; 2: 113–120.
8. Lee SM, Suh DH, Kim SY, et al. Antenatal Prediction of Neonatal Survival in Sacrococcygeal Teratoma. J Ultrasound Med. 2018; 37(8): 2003–9. https://doi.org/10.1002/jum.14553
9. Graf JL, Albanese CT. Fetal sacrococcygeal teratoma. World J Surg. 2003; 27: 84–6. https://doi.org/10.1007/s00268-002-6741-6.
10. Westerburg B, Feldstein VA, Sandberg PL, et al. Sonographic prognostic factors in fetuses

with sacrococcygeal teratoma. J Pediatr Surg. 2000; 35: 322–6. https://doi.org/10.1016/s0022-3468(00)90032-0

11. Grigore M, Iliev G. Diagnosis of sacrococcygeal teratoma using two and three-dimensional ultrasonography: two cases reported and a literature review. Med Ultrason. 2014; 16(3): 274–7. https://doi.org/10.11152/mu.2013.2066.163.mg1gi2
12. Usui N, Kitano Y, Sago H, et al. Outcomes of prenatally diagnosed sacrococcygeal teratomas: the results of a Japanese nationwide survey. J Pediatr Surg. 2012; 47(3): 441–7. https://doi.org/10.1016/j.jpedsurg.2011.08.020
13. Neubert S, Trautmann K, Tanner B, et al. Sonographic prognostic factors in prenatal diagnosis of SCT. Fetal Diagn Ther. 2004; 19: 319–26. https://doi.org/10.1159/000077959
14. Wilson RD, Hedrick H, Flake A, et al. Sacrococcygeal Teratomas: Prenatal Surveillance, Growth and Pregnancy Outcome. Fetal Diagn Ther. 2009; 25: 15-20.
15. Mieghem VT, Al-Ibrahim A, Deprest J, et al. Minimally invasive therapy for fetal sacrococcygeal teratoma: case series and systematic review of the literature. Ultrasound Obstet Gynecol. 2014; 43(6): 611–9. https://doi.org/10.1002/uog.13315
16. Wang Y, Wu Y, Wang L, Yuan X, Jiang M, Li Y. Analysis of Recurrent Sacrococcygeal Teratoma in Children: Clinical Features, Relapse Risks, and Anorectal Functional Sequelae. Med Sci Monit. 2017; 23:17-23.