

Sacrococcygeal Tumor – A Rare Case Report

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ABSTRACT

Sacrococcygeal teratoma (SCT) is a rare congenital tumor primarily affecting infants, with a prevalence of approximately 1 in 35,000 live births. These tumors, often detected either prenatally or shortly after birth, originate from germ cells and typically occur at the base of the coccyx. Despite their benign nature, SCTs can pose significant risks to fetal and maternal health, necessitating careful management and monitoring. A 32-year-old primiparous woman presented to Siak Regional Hospital at 36 weeks of gestation with complaints of uterine contractions and bloody mucus discharge from the birth canal. A physical examination revealed a palpable mass in the uterine fundus and pubic symphysis. Ultrasonography confirmed the presence of a fetal SCT with a solid mass measuring 6.2 x 8.1 x 7.3 cm in the sacral region, and a cesarean section was performed. The infant, a baby girl, was born with a birth weight of 2150 grams and an Apgar score of 3/4. Congenital abnormalities were observed in the form of a solid mass around the sacrum, indicative of SCT. SCT poses challenges in prenatal diagnosis, management, and prognosis. Prompt recognition and multidisciplinary management are essential for optimizing outcomes. Further research is needed to enhance understanding and improve therapeutic strategies for this rare congenital anomaly. SCT is a rare congenital anomaly that requires early diagnosis. Thorough examination with ultrasonography is essential to rule out other congenital anomaly, and predict prognosis.

Keywords: cesarean section delivery, infantile congenital tumor, multidisciplinary management, prenatal diagnosis, sacrococcygeal teratoma

Sacrococcygeal teratoma (SCT) is a rare tumor typically found in infants, often diagnosed either before or shortly after birth. These tumors, mainly mature teratomas, share similarities with germ cell tumors (GCTs) but are distinct entities. Although usually noncancerous, there's a possibility of recurrence or transformation into other tumor types.^{1,2} Patients with SCT often encounter gastrointestinal, urological, and neurological issues, affecting their quality of life. Due to its rarity and absence of standardized management, numerous clinical questions remain unanswered. Insights from developmental biology and genetics have provided clarity on its origin and development.³ Diagnosis typically involves elevated alpha-fetoprotein levels or ultrasound findings. While surgery is generally successful, larger tumors may necessitate extensive

intervention and monitoring, particularly if they pose fetal health risks.^{4,5}

CASE PRESENTATION

A 32-year-old primiparous woman came to Siak Regional Hospital at 36 weeks of gestation with complaints of uterine contractions for 2 hours before entering the hospital. Complaints include bloody mucus discharge from the birth canal. The patient denied any history of amniotic fluid or blood discharge. On Leopold's examination, a hard, round mass was palpable in the uterine fundus and pubic symphysis. The patient only had one ANC visit at 2 months of pregnancy and had never had an ultrasound examination. When we did an ultrasound examination, we found a fetus with a breech presentation with an estimated fetal weight of 2200 grams, a fetal heart rate of 145 bpm, a visible mass measuring 6.2 x 8.1 x 7.3 cm in the sacrum of the fetus with visible neovascularization, and the impression of a sacrococcygeal Due to the

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difficulty of having a vaginal birth, we decided to do a cesarean section.

Then, a baby girl was born with a birth weight of 2150 grams and an Apgar score of 3/4. Congenital abnormalities appear in the form of a solid mass around the sacrum, the size of a baby's head. The baby was treated in the perinatology department and planned for a consultation with a surgeon, then the surgeon examined him and planned an MRI examination to determine the next course of action, but the baby died one day later due to respiratory failure. During this pregnancy, the patient admitted that she had never had a history of illness, and there was no history of birth defects in the patient's family or the patient's husband.

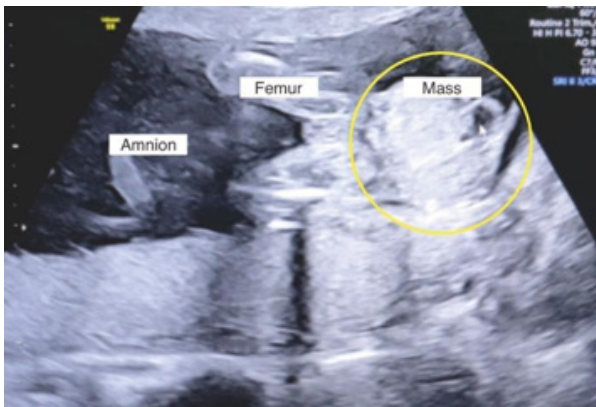


Fig1. Ultrasound examination shows a mass in the sacrum of the fetus (yellow circle).

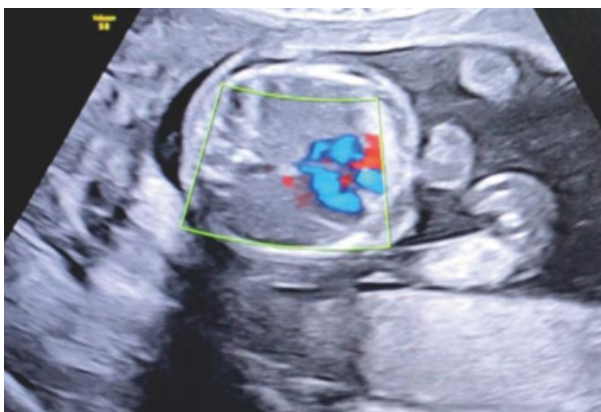


Fig 2. Neovascularization was visible in the mass from the Doppler ultrasound results.



Fig 3. Photo of the baby after birth, showing a solid mass on the sacrum.

DISCUSSION

Sacrococcygeal tumor is a common germ cell tumor found in 1:35,000 fetuses. Specifically, it was three to nine times more likely to be found on female fetuses. The tumor arises in the midline pre-sacral area and can extend to the pelvis and abdominal cavity.^{6,7} The American Academy of Paediatrics classifies the tumor into 4 classes: major external mass (Type 1), external with intra-pelvic extension (Type 2), internal with visible external extension (Type 3), and internal with minimal external extension (Type 4). The case presented is type 1.^{8,9} Sacrococcygeal tumors is suspected to grow from primitive cells during embryogenesis. Hansen Node is a pluripotent cell that can develop into embryonic and extra-embryonic tumor. tumors. The abnormal growth will shows mostly in midline area. Another hypothesis also mentions that the tumor might grow from embryonic cells.¹⁰

Ultrasound, especially during the first trimester, is very essential for early detection. However, our patient did not undergo prenatal examinations regularly, which might explain the late detection of the anomaly.^{11,12}

Sacrococcygeal tumors usually cause prenatal contractions due to their common co-occurrence with polyhydramnios. A large tumor might cause uterine retention, disrupting amniotic fluid volume regulation.^{10,12} High metabolic demand from tumor growth might also cause anemia. Amnioreduction

is usually performed to prevent preterm labor.¹³ Other prenatal interventions might involve vascular shunt surgery or in-utero tumor resection (through radio-ablation or open fetal surgery).¹⁴ No prenatal intervention was done in this patient due to the late finding.

The Japanese Sacrococcygeal Teratoma Guideline recommends a cesarean section for a lesion outside the pelvic to avoid rupture, haemorrhage, and delivery complications.⁹ With the consideration of a big extra pelvic tumor, we perform a cesarean section. No labor or delivery complications were found in this case.¹⁵

This congenital anomaly has a bad prognosis. Fetal deaths occur in almost all fetuses with hydrops because this anomaly is usually also found with arterial-venous (AV) fistulas, causing cardiac failure.^{3,10} This might explain the fetal death in our patient; however, we do not perform an autopsy to confirm the AV fistula hypothesis. A better prognosis is found in patients with hydrops, where research found that almost all non-hydropic fetuses ended up surviving. In this case, the impression is that the baby does not have fetal hydrops.^{13,16}

CONCLUSION

Sacrococcygeal tumor is a rare congenital anomaly that requires early diagnosis. Thorough examination with ultrasonography is essential to rule out other congenital anomaly, and predict prognosis. Late diagnosis and intervention can resulted in fetal death.

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