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Successful management of pregnancy with fetal *sacrococcygeal teratoma* by multi-dicsciplinary approach: A case report

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Abstract
Sacrococcygeal teratoma (SCT) is a rare germ cell neoplasm in the newborn with a higher incidence rate in female baby. SCT are diagnosed on ultrasound as exophytic masses rapidly growing in utero. Most of *Sacrococcygeal teratoma* are non-cancerous and increasing metabolic demands of the rapid but disproportionately growing tumor results in high risk of perinatal and obstetric complications.
At birth, SCT dystocia, intra tumoral haemorrhage or tumoral rupture are of major concern and are life threatening. Timely management becomes the key for a good perinatal outcome.
A 32-year-old, multi gravida mother visited us for regular antenatal checkups. Her routine blood investigations were normal. Anomaly scan was done at 19 weeks of gestation showed a 24 cc exophytic mass arising from the sacrum with no obvious involvement of spinal canal. No other associated malformations noted. The Tumor volume: Fetal Weight Ratio (TFR) was 0.069, which suggested a good fetal outcome.
The risks and complications of continuing pregnancy were explained to the couple & they opted to continue.

Keywords: *Sacrococcygeal teratoma* (SCT), germ neoplasm, exophytic mass

Introduction
Sacrococcygeal teratoma (SCT) is a rare germ cell neoplasm in the newborn. It develops before a baby is born and grows from tailbone, with an incidence of 1 in 20000-40000 live births. There is 3-4-fold increase incidence in female baby [1].
It can grow to a massive size in utero. Most of *Sacrococcygeal teratoma* are non-cancerous and increasing metabolic demands of the rapid but disproportionately growing tumor results in high risk of perinatal complications, fetal anemia, intra tumoral haemorrhage, intrauterine fetal demise due to non-immune hydrops, high output cardiac failure and obstetric complications such as preterm labor, polyhydramnios, obstructed labour. SCT may induce maternal mirror syndrome characterized by edema and hypertension.
At birth, SCT dystocia, intra tumoral haemorrhage or tumoral rupture are of major concern and are life threatening. SCT are diagnosed on ultrasound as exophytic masses rapidly growing in utero, and timely management becomes the key for a good perinatal outcome [2-4].

Case Report
Here we present the case of a 32-year-old, G2A1 expecting mother. She had a molar pregnancy in Jan 2021 for which suction evacuation was done. HPE report was conclusive of a benign mole. She is a known case of hypothyroidism on medication. There is no significant family history.
The present pregnancy was confirmed by ultrasound, gestation corresponded with dates. She received regular immunization of pregnancy and was on antenatal supplements. All antenatal investigations including double marker and NT Scan were normal.
At 19 weeks anomaly scan showed a 24 cc exophytic mass arising from the sacrum with 20 cc external and 4 cc internal component, with 80% solid and 20% cystic component, with no obvious involvement of spinal canal. No other associated malformations. No secondary mass effect/hydrops/polyhydramnios no intratumoral haemorrhage.

The Tumor volume: Fetal weight ratio (TFR) was 0.069 which suggested a good fetal outcome. Fetal MRI confirmed the diagnosis. The lesion measured 2.5/2.3 cm on MRI.

Couple opted to continue pregnancy. The risks and complications of continuing pregnancy were explained to the couple.

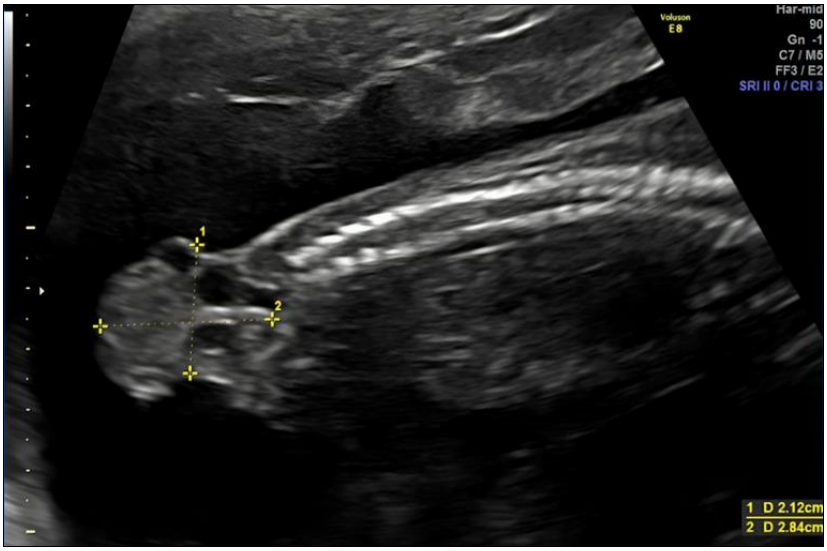

Fig 1: Anomaly scan: 24 cc exophytic mass arising from the sacrum with no obvious involvement of spinal canal

At 23 weeks, pediatric surgeon opined conservative therapy. At 26 weeks, there was no increase in the size of the tumor.

At 29 weeks, growth scan revealed polyhydramnios with AFI 40, and the lesion measured 9x7x8 cm with volume of 296 cc with solid: Cystic components of 70:30%. Because of the rapid growth in the tumor, the decision to deliver was taken. Antenatal corticosteroids were given for fetal lung maturity. Neonatal counselling given and posted for elective LSCS.

At 30 weeks 3 days, elective LSCS was done. Intraoperatively, the lower uterine segment was stretched, very vascular with engorged blood vessels. Liquor was thin meconium stained. A female baby weighing 2.48 kg was delivered by vertex. Baby had *sacrococcygeal teratoma* of about 15 x 10 cm with multiple surface ulcerations. Baby had good cry at birth and was handed over to pediatrician for neonatal care.

Patient had atonic PPH, managed with Injection Carbetocin, intramyometrial carboprost, oxytocin and 800 mcg of Misoprostol. Blood loss was around 800ml. Patient did not require any blood transfusion. Post-operative period was uneventful and patient was discharged on the third day of caesarean section.

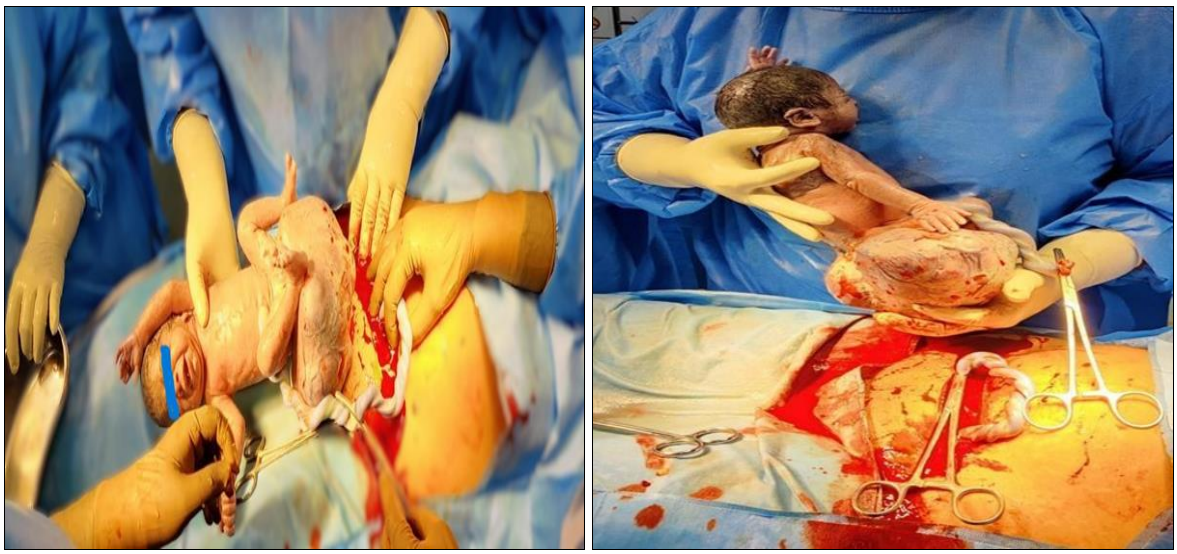

Fig 2 & 3: At Elective LSCS, baby weighing 2.48 kg with SCT

Meanwhile, baby was admitted in NICU. On D3, baby underwent surgical excision of the tumor. On table, baby had cardiac arrest, revived by pediatric anesthetist. Baby had massive blood loss. 3 PRBC were transfused. Baby was put on ventilator for 10 days. Baby had respiratory distress syndrome, Patent ductus arteriosus, sepsis, hemorrhagic shock, Broncho pulmonary dysplasia, feed intolerance with gastro esophageal reflux disorder, apnea of prematurity, neonatal hyperbilirubinemia. Baby received appropriate neonatal intensive care and was discharged on 53rd day of delivery

Histopathology of tumor mass confirmed Immature Teratoma

Fig 3: Baby at level II B NICU

Baby is under regular monitoring with Paediatrician and paediatric surgeon with routine ultrasound and alphafetoprotein. Baby is doing excellent with no signs and symptoms of recurrence. At the age of 1.2 years baby started walking and there is no neurological deficits or incontinence. Developmental milestones achieved till date without any developmental delay.

Discussion

Sacrococcygeal teratoma is derived from the remnants of the primitive streak. Almost 75% of SCT are benign, 12% are malignant and the remaining are immature teratomas that share benign and malignant features. Anatomically, Altman *et al* classified SCT depending on the degree of intra- and extra pelvic components. Type I is primarily external, type II is primarily external but with significant intra-pelvic component, type III is mainly intra-pelvic with small external component and type IV is completely intra-pelvic with no external components. Type I and II usually present as a visible lump or mass in the sacral region beneath the skin at the top of the buttocks crease. Type III and IV may not be visible and just felt on palpation.

Prenatal diagnosis can be made by ultrasonography as early as 13 weeks of GA. The tumor growth and occurrence of complications are detected by regular ultrasound evaluation.

A tumoral size greater than 4 cm at a GA of 20 weeks warrants a weekly US monitoring. Serial ultrasound monitoring should be done to evaluate the tumor size, the solid or cystic portion, and the amount of amniotic fluid, as well as the vascular flow using Doppler. High-risk tumors are the ones that are larger or with a rapid growth, mostly solid, and with high vascular flow. They can create a vascular steal phenomenon that can cause cardiomegaly and hydrops [5].

Magnetic resonance imaging (MRI) helps in differentiating SCT from distal neural tube defects, which is an important differential diagnosis. The main difference is the location of the mass effect, which is presacral in SCTs and posterior in neural tube defects. It allows an accurate assessment of the extension of tumor, presence/ absence of the internal component and the intra-pelvic/ intra-abdominal/ intraspinal extension, compression of adjacent organs, the extent of solid and cystic areas within the tumor and the presence of intra-tumoral haemorrhage. MRI helps to make an improved preoperative plan for surgical resection.

Pregnancies with fetal SCT develop several complications due to the rapidly increasing size of the tumor. It can result in fetal decompensation and intra uterine fetal demise, due to high output cardiac failure, severe fetal anemia due to intra tumoral haemorrhage, and non-immune hydrops. Obstetric complications such as preterm labour, polyhydramnios and PPROM are commonly seen. SCT may induce maternal mirror syndrome characterized by maternal edema and hypertension. At birth, there can be labour dystocia especially with larger tumors, intra tumoral haemorrhage or tumoral rupture can happen during vaginal delivery and are of major concern as they are life threatening.

Factors associated with an increased morbidity and mortality include tumor size larger than 10 cm, tumor volume to fetal weight ratio (TFR) > 0.12 prior to 24 weeks of gestation, rapid growth rate higher than 150 cubic cm/week, the presence of solid components, increased vascularity, hemorrhage, tumor rupture, fetal hydrops, high-output cardiac failure, polyhydramnios. Factors associated with a better prognosis include small (< 7 cm) and poorly vascularized cystic masses [5-8].

An accurate diagnostic assessment by USG and MRI enables us to categorize the patient risk-wise and plan further management in terms of the time and mode of delivery, prenatal or postnatal intervention of the tumor.

A prenatal intervention is considered in a fetus < 28–30 weeks at high-risk for preterm delivery and fetal demise due to high-output cardiac failure if left untreated. Intrauterine intervention can be made by laser ablation of feeding vessels.

An emergency cesarean delivery should be preferred at > 30 weeks of gestation in patients at high risk for preterm delivery or fetal demise [9]. An elective cesarean delivery should further be preferred in SCT > 5 cm in diameter or > 750 cm [7] in volume to avoid dystocia, tumor rupture and hemorrhage [7, 9]. In cases not at high-risk for preterm delivery and fetal demise pregnancy continuation and term vaginal delivery are feasible.

Conclusion

In our case, poor prognostic factors such as tumor size >10 cm, high TFR, rapid tumor growth rate, increase in the solid to cystic ratio, increase in the vascularity and polyhydramnios started to

appear. Timely detection of these features was possible due to regular and accurate imaging studies. An immediate delivery after antenatal corticosteroid coverage was planned as the fetus was certainly heading fast towards decompensation. In a tertiary center with good NICU facilities, availability of pediatric surgical and anesthetic team, early delivery before the decompensation sets in would be prudent for a favorable outcome. All the above-mentioned factors must be taken into consideration to individualize the treatment plan.

Conflict of Interest
Not available

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Not available

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