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A RARE CASE OF SACROCOCCYGEAL TERATOMA: PRENATAL DIAGNOSIS AND MANAGEMENT

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ABSTRACT

BACKGROUND- The aim of this paper is to present a rare case of sacrococcygeal teratoma in fetus.

CASE REPORT- A 25-year-old primigravida was referred to Government Medical College, Nagpur with sonographic finding of SLIUG of 38 weeks of gestation with sacrococcygeal mass of 10*10 cm. Patient was in labor when she reached at hospital. ANC ultrasound done at 20 weeks of gestation was within normal limit and there was no evidence of any sacrococcygeal mass or any other anomalies. USG done at 38 weeks showed a large heterogenous well defined, hyperechoic structure arising from sacrococcygeal region of fetus of size 8.2*7.9*8.3 cm with multiple hyperechoic areas seen within showing increased vascularity, features suggestive of sacrococcygeal teratoma. Considering the size of tumor, patient was taken for emergency caesarean section and delivered a male baby of 3.5 kgs. Baby cried immediately after birth. APGAR at 5 minutes was 9. A large 10*10*8 cm mass was seen arising from sacrococcygeal region, showing dilated veins over the surface. Baby was shifted to NICU for monitoring. On day 3 the baby was posted for sacrococcygeal teratoma excision. Complete surgical resection of the tumor done.

CONCLUSION- Sacrococcygeal teratoma though rare is the most common tumor in the neonatal period. Prompt diagnosis and elective cesarean section reduce the maternal and fetal complications. A multidisciplinary team approach is cornerstone in successful management of neonates with sacrococcygeal teratomas.

KEYWORDS : sacrococcygeal teratoma, mature teratoma

INTRODUCTION

The incidence of sacrococcygeal teratoma (SCT) is 1:35000 to 1:40000 live births[1]. It is the commonest congenital tumor in neonates. Teratoma originates from the early embryonic pluripotent stem cells and the Hansen's node in front of the coccyx is the site where the pluripotent stem cells are concentrated[2]. SCTs are comprised of different types of tissues that come from at least two of the three germ cell layers. Depending on the tissues that are included, they are divided into mature, immature or malignant teratomas. It is more common in females with a male: female ratio of about 1:3-4[3]. Based on external component and intra pelvic/intra abdominal extension of the tumor, Altman classified SCT into 4 types. (American Academy of Pediatrics Surgical Section Classification)[4]. SCTs seen at birth are usually Altman Type 1 and 2 (87%)[5]. Rarely type 3 can also be seen in neonates, while type 4 is typically seen later in life with no external component. There is a high risk of perinatal complications and neonatal mortality due to maternal obstetric complications like tumor rupture, preterm labor, or dystocia. The fetus is also at risk of high output cardiac failure, placentomegaly and hydrops with subsequent fetal demise secondary to increased metabolic demands and vascular steal of a rapidly growing solid tumor[6]. Additionally, hydrops can cause "maternal mirror syndrome" that is life threatening for the mother. It is worth noting that prognosis has nothing to do with the size of the tumor. However the prognosis of parenchymal vascular masses is worse than that of cystic masses. Modern technology like three-dimensional sonography can be used for early diagnosis even in the first trimester. In this report we describe a case of SCT diagnosed late in pregnancy and managed successfully postnatally.

Case Presentation

A 25-year-old primigravida was referred to Government Medical College, Nagpur with outside sonographic finding of single live intrauterine gestation of 38 weeks of gestation with sacrococcygeal mass of 10*10 cm. The patient had no family history of birth defects or genetic disorders. She was married since 1 year. It was a non consanguineous marriage. She was a booked case at private hospital with regular ANC visits. ANC ultrasound done at 20 weeks of gestation was within normal limit and there was no evidence of any sacrococcygeal mass or any other anomalies. Her oral glucose challenge test was within normal limit. She was in labor when she reached hospital. She was admitted in labor ward. Her relatives were thoroughly counseled about the need for cesarean section, possible NICU admission of the baby and the need for further surgery on the baby to remove the tumor.

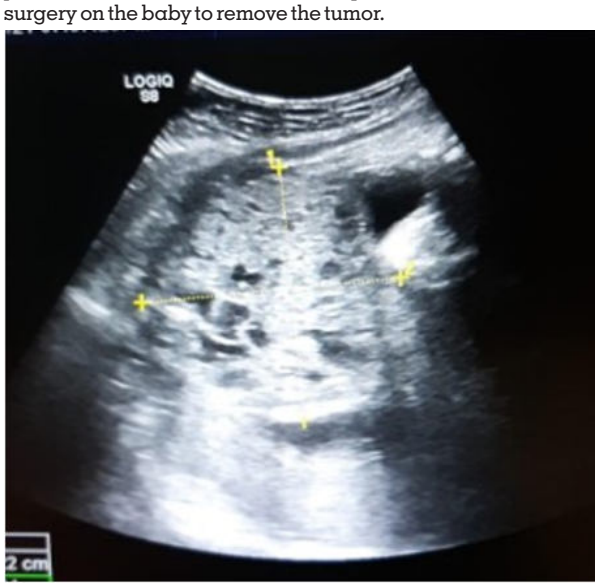

Fig. 1. ANC USG showing sacral mass in fetus

On examination, her vitals were stable, pulse =86/min, blood pressure =120/80 mmHg. Mild pallor was present. On per abdominal examination, uterus was full term, not overdistended cephalic presentation, head was floating, liquor was clinically adequate, fetal heart sounds were heard at left occipital transverse position, uterus was relaxed. On per

vaginal examination, cervix was 3cm dilated, cervical length was 2 cm, mid placed, effacement was 30-40%, membranes was present, vertex presentation, station was above brim, pelvis was adequate. A sonography was done from our Institute to confirm the diagnosis of sacrococcygeal teratoma. USG showed a large heterogenous well defined, hyperechoic structure arising from sacrococcygeal region of fetus of size 8.2*7.9*8.3 cm with multiple hyperechoic areas seen within showing increased vascularity, features suggestive of sacrococcygeal teratoma. Considering the size of tumor, patient underwent an emergency caesarean section and delivered a male baby of 3.5 kgs. Baby cried immediately after birth. APGAR at 5 minutes was 9. A large 10*10*10 cm mass was seen arising from sacrococcygeal region, showing dilated veins over the surface. Baby was shifted to NICU for monitoring. USG abdomen of baby was within normal limit. USG spine showed no evidence of any outpouching from the spinal canal (Cervical-lumbar). No obvious bony deformity noted. USG local- evidence of large, well defined solid cystic lesion in the sacral area of approx. size 14*10*12cm with multiple cystic foci within taking mild vascularity, features suggestive of neoplastic etiology likely sacrococcygeal teratoma. On day 3 the baby was posted for SCT excision. Complete surgical resection of the tumor wall. The levator sling was well preserved. The baby was shifted to NICU postoperatively for monitoring and management. Baby discharged from NICU on day 12. Now baby is normal and with mother. Histopathology of excised tumor mass suggestive of mature teratoma (tumor shows squamous epithelium, glandular areas, mature cartilage, mature neural tissue admixed together).

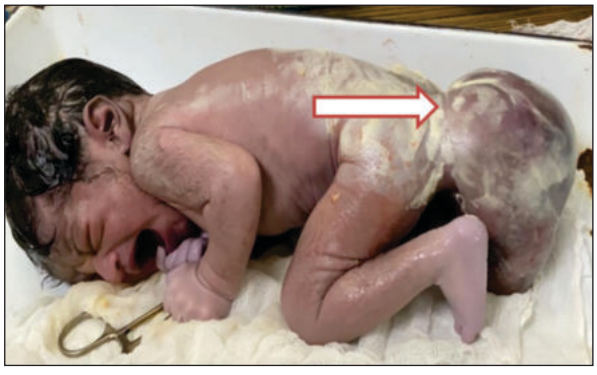

Fig. 2. Sacrococcygeal mass of newborn

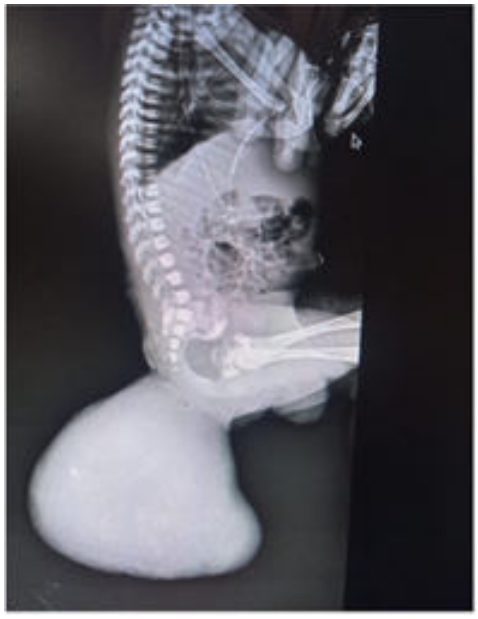

Fig. 3. X-ray lateral view

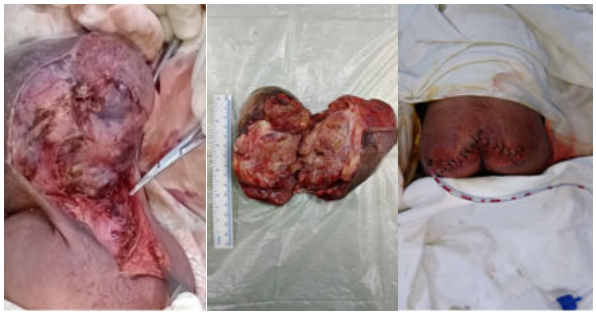

Fig. 4. Intra operative Fig. 5. Cut section Fig. 6. Post-operative

Fig. 7. 1 month infant for follow-up

DISCUSSION

The term "teratoma" is derived from the Greek word "Teraton" which means monster[7]. Sacrococcygeal teratomas are classified according to American Academy of Pediatrics Surgical Section[5].

Type 1- The tumor is predominantly external with a very minimal internal component. Type 1 is rarely associated with malignancy.

Type 2-The tumor is predominantly external but has some internal extension into the presacral space.

Type 3- The tumor is visible externally but is predominantly located in the pelvic area with some extension into the abdomen.

Type 4-The tumor is not visible externally and is in the presacral space. It has the highest rate of malignancy.

The prognosis of the newborn with SCT depends on the time of diagnosis, malignant potential of the tumor and the ease of surgical excision[8]. Prenatal ultrasonographic examination is useful in the diagnosis of SCTs and the main manifestation is sacral mass. Early prenatal presentation is associated with increased morbidity and mortality while diagnosis after 30 weeks of gestational age is a good prognostic indicator for fetal survival. MRI can also be done for diagnosis of SCTs. In our case the diagnosis was made at 39 weeks indicating better fetal outcome and MRI could not be done as patient had labor pains. Also, our case is that of a male newborn. Although 75% SCTs are seen in females, the exact reason for female preponderance is not exactly known[9].

Alfa fetoprotein can be used as a marker to differentiate between mature and malignant teratomas at presentation and during follow up of the patients.

Cesarean section is suggested when the diameter of fetal SCTs is more than 5 cm to avoid dystocia, birth injury, intertumoral hemorrhage and tumor rupture[10]. As the tumor in our case was 10cm we proceeded with cesarean section.

Some fetal SCTs are highly vascular and grow rapidly which results in polyhydramnios, fetal blood loss, high output fetal

cardiac failure, fetal anemia, fetal edema, and death. SCTs can also result in fetal bladder obstruction, fetal hydronephrosis and fetal ureteral dilatation[10].

Fetal intervention can be done to manage the complication of SCTs which includes open fetal surgery to resect the tumors, radiofrequency ablation, major vessel laser ablation and vessel alcohol sclerosis to prevent high output fetal cardiac failure and reduce intra tumor arteriovenous shunt. It also reduces tumor volume and fetal oedema thereby improving fetal outcome. Amnioreduction and cyst aspiration can also be done to prevent preterm labor and tumor rupture at delivery[11].

The percentage of malignant transformation within 2 months after birth is 20% and 40% after 4 months, so SCTs should be completely removed as soon as possible after birth to prevent malignant transformation. Hence we removed the tumor as soon as possible. Predominant risk factors for tumor recurrence are residual coccyx and tumor rupture during operation[12]. So, tumor and coccyx should be removed as completely as possible. At present most professionals thought that for benign teratoma, removing the coccyx and avoiding damage to the residual cyst wall during surgery is the key to prevent recurrence. Other studies have shown that even after complete resection SCTs can recur many years after initial resection. So close follow up till adulthood is necessary[12].

CONCLUSION

Sacrococcygeal teratoma though rare is the most common tumor in the neonatal period. Prompt diagnosis and elective cesarean section reduce the maternal and fetal complications. Early excision of tumor and meticulous follow up yields good results. Thus, a multidisciplinary team approach involving obstetricians, fetal medicine specialists, radiologists, neonatologists, pediatric surgeons, anesthesiologists, intensivists form the cornerstone in successful management of neonates with SCTs.

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