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Case Report

Antenatally undetected giant sacrococcygeal teratoma in a neonate: a case report with review of literature

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ABSTRACT

Sacrococcygeal teratomas are the most common germ cell tumors in neonates, often diagnosed antenatally. Antenatal detection can guide delivery planning and early intervention. We report a case of a female neonate born with a giant sacrococcygeal teratoma that was missed on four antenatal ultrasounds, including a level II scan. Delivered vaginally at term, the neonate underwent successful surgical excision on day 3 of life, with no neurological or urogenital sequelae. This case highlights the critical role of thorough prenatal imaging in detecting sacrococcygeal teratomas, enabling early intervention and favourable outcomes.

Keywords: Sacrococcygeal teratoma, Prenatal diagnosis, Neonatal surgery

INTRODUCTION

Sacrococcygeal teratoma is a type of non-organ-specific extragonadal germ-cell tumour that occurs at the base of the coccyx and can be detected prenatally or at birth. Despite its rare occurrence, It is the most common type of germ cell tumour in neonates and infants with an incidence of 1 in 35,000 to 40,000 live births and has a predilection towards the female sex (3:1 ratio).[1] They can present as benign (mature) or malignant (immature) tumours.[2] Rapid growth in size and high vascularity of these tumours can cause mass effect, haemorrhage, and hemodynamic compromises during pregnancy and can lead to high output cardiac failure and fetal hydrops which is considered a poor prognostic marker. These Prenatal and perinatal complications require optimal obstetric and surgical management.[3] Routine fetal anomaly screening helps in prenatal diagnosis of these tumours. Fetal ultrasound with Doppler imaging and more recently fetal MRI may be used to document the extent of the tumour as well as identify the population at risk for serious fetal complications. Vaginal delivery is considered if the size of the tumour is small and for large tumours >5 cm size usually elective caesarean section is planned.[4] This case report showcases the successful resection of the antenatally undetected tumour, in a baby delivered vaginally; fortunately, without any fetal and intrapartum complications.

CASE REPORT

This female newborn, born to a 26-year-old G2P1L1 mother at a gestation of 39+4 weeks, with a birth weight of 3.25 kg was referred to us by an obstetrician on day 2 for life with the finding of a large mass observed at birth in coccygeal area of the baby. There was no significant antenatal history from the mother. It was a booked and supervised uneventful pregnancy with adequate prenatal care from the commencement. Mother had undergone a total of 4 antenatal ultrasounds including one level II anomaly scan and all four were grossly normal. There was no ultrasound in the last trimester. As all four ultrasounds were grossly normal, the obstetrician didn't anticipate any intrapartum complication and gave a trial of normal vaginal delivery. The baby was born via vaginal delivery and at birth the mass was observed by the obstetrician and the baby was referred to our centre on day two of life.

Physical examination

The baby was vitally stable on admission. Grossly normal head size and shape; except for the sacrococcygeal mass, no dysmorphism or gross malformation was observed on head-to-toe examination. On systemic examination also a normal cardiopulmonary system; soft abdomen, no organomegaly, and no central nervous system involvement were seen. The baby's higher mental functions, tone, activity, reflexes were normal as per gestation age. The mass was externally visible, protruding from the lower back, and with a dimension of 14×10×6 cm^3 with the anal opening displaced anteriorly. It appeared cystic with areas of firmness and discolouration (Figure 1).

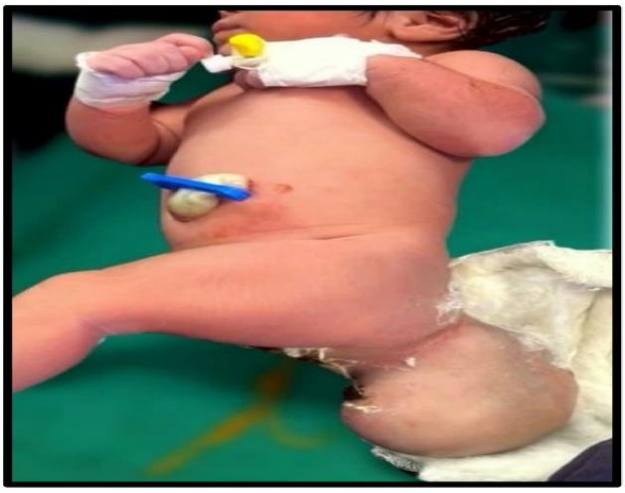

Figure 1: Female neonate with sacrococcygeal teratoma.

Management and outcome

The baby was admitted and started on I. V. antibiotics and routine investigations were sent which came out to be within normal range. X-ray showed a cystic mass with calcifications visible at the base of the mass (Figure 2). Ultrasound imaging of the mass along with the abdomen and pelvis revealed a mixed echogenic mass with cystic and solid components originating from the sacrococcygeal region, consistent with a sacrococcygeal teratoma. The internal organs appeared normal, with no signs of metastasis.

A multidisciplinary team, including a neonatologist, paediatric surgeon, and anaesthesiologist, decided to proceed with early surgical intervention given the size of the tumour and potential complications. The neonate underwent surgery on day 3 of life with complete resection of the tumour, including the coccyx to reduce the risk of recurrence. Careful dissection to avoid damage to the surrounding structures was performed (Figure 3). Histopathological examination of the resected tumour confirmed it to be a mature teratoma, predominantly cystic with no malignant components.

Postoperatively the neonate was closely monitored in the neonatal intensive care unit (NICU) for signs of complications such as bleeding, infection, and bowel or bladder dysfunction. For pain management baby was provided with appropriate analgesia. Feeds were started 6 hours after surgery and gradually hiked to full breastfeeding. The drain was kept for 5 days and then removed and the baby was discharged on day 7 post-surgery in a vitally stable condition with normal systemic examination and full bladder and bowel functions and no motor deficits in any limb. Parents were advised to follow up in OPD for follow-up USG/MRI and to look for recurrence.

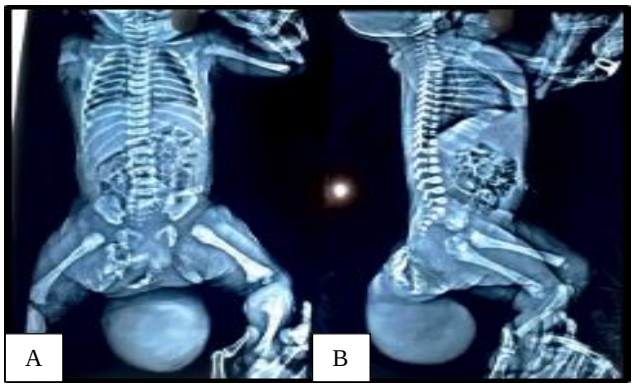

Figure 2 (A and B): Infantogram showing cystic mass with calcifications (white arrows).

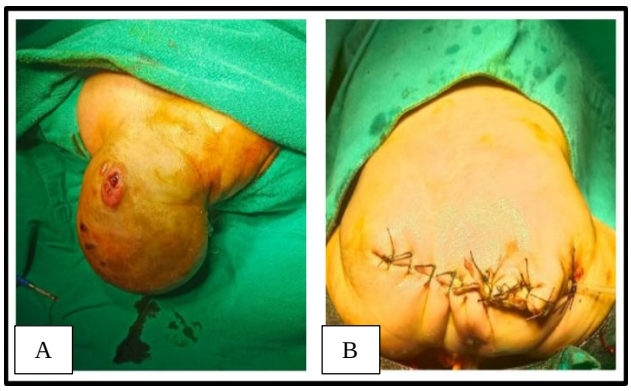

Figure 3: Pre (A) and post-surgery (B).

DISCUSSION

Sacrococcygeal teratomas are extragonadal neoplasms originating from the presacral area. The incidence of Sacrococcygeal teratomas is 1/40,000 live births and a prevalence of 1/21700 live births. It is the most common germ cell tumour in neonates and infants <2 years of age. These tumours usually present as a midline large cystic or solid mass. The most common location is the sacrococcygeal area followed by gonads, retroperitoneum, cervix, mediastinum and oropharynx. Gross cytogenetic/chromosomal abnormalities have not yet been established with these tumours and mostly are sporadic with no familial tendencies but in 10-15% of

cases, family history of twins has been observed. It also has a sex predilection towards females, but the recurrence rate is higher in males.[5-8]

These tumours contain various kinds of body tissues which are foreign to the site of their origin. They can be benign or malignant, cystic or solid and mostly present in the neonatal period. SCT can grow enormously in size, causing mass effect, bladder outlet obstruction and dystocia.[6,9]

The SCT has been classified into 4 types (Altman classification) according to their location. The most common type is the type I SCT (45%) which is mostly external, with minimum intrapelvic component, and has the least malignant potential. Type II SCT (35%)-is predominantly external with a significant intrapelvic component. Type III SCT (10%)-is predominantly intrapelvic, with visible external extension. Type IV SCT (10%)-is intrapelvic with possible visible external part and is the most malignant type.[3]

Histologically SCT can be differentiated into three types. 1. Mature type-with well-differentiated tissues. 2. Immature type- along with mature tissues, they contain neural tube-like structures and is further classified into four types depending on the amount of immature tissue present. 3. Teratoma with malignant components: contains one or more malignant germ cell tumours.[10,11]

Most neonates with SCT are asymptomatic at birth, but large tumours can cause bowel or bladder dysfunction. 15-30% of SCTs are associated with congenital anomalies, with urogenital system anomalies such as hydronephrosis being the most common. Other malformations can also be associated such as hip dislocation, spinal dysraphism, and cardiac anomalies.[12]

Approximately 25-50% of SCTs are diagnosed in utero during routine USG screening.[13,14] Diagnosing SCT in the first trimester of pregnancy or the second-trimester screening helps in the morphological classification of SCT, assessment of the tumour size, growth, percentage of solid component, vascularization, and associated congenital defects and would improve the prognosis of these cases and allow early postnatal surgical intervention.[15] USG can also be used to monitor tumour progression, detect complications and plan the management.[16]

Sacrococcygeal teratomas are associated with morbidities and mortality from complications like preterm birth, malignant invasion, haemorrhage, umbilical flow obstruction and high-output heart failure.[17] Fetal demise is seen with solid and highly vascularized fast-growing tumours leading to high output cardiac failure and fetal hydrops.[18] Some of these complications can be detected prenatally and are treatable. That makes the antenatal diagnosis of SCT very important.

Large SCTs can lead to intrapartum complications like dystocia, tumour rupture, birth asphyxia, PPH. However, babies with a small SCT (<5 cm) can be safely delivered vaginally, for large SCTs elective caesarean section should be done.[4] Older studies have reported a high rate of caesarean section (75%), while recent studies have indicated that 43 out of 44 neonates with SCT were successfully vaginally delivered.[19,20]

The mainstay of management of SCT is en bloc resection of the tumour irrespective of the histological type.[21] Delayed treatment may lead to tumour rupture and haemorrhage. The surgical outcome and prognosis of SCT are favourable. The recurrence rate after surgery has been estimated as 10-15%. Incomplete resection and immature/malignant histology are the most important risk factors for recurrence.[22] Although immature teratoma histology is a risk factor for recurrence, postoperative chemotherapy is not recommended because it is ineffective in preventing recurrence.[23] In addition to classic surgical resection for SCT, some innovative in-utero interventions to rescue complicated fetuses with a large SCT or to decrease surgical bleeding in neonates are still investigational.

Regular follow-up examinations are performed by imaging procedures and tumour marker tests of alpha-fetoprotein. There is little evidence to provide guidance on follow-up care for children with sacrococcygeal teratomas.

CONCLUSION

The case reported here creates awareness regarding the importance of antenatal detection of sacrococcygeal teratoma for anticipation of possible hemodynamic compromises during pregnancy and their management. The identification of high-risk cases through the imaging evaluation of unfavourable prognostic factors like percentage of solid components, rapid growth in size, and hypervascularity are of utmost importance. Also, it will help the obstetrician as well as the neonatologist in planning the best possible perinatal and post-natal management of such babies for better outcomes. Multidisciplinary collaboration is crucial for optimizing outcomes, and ongoing follow-up is necessary to monitor for potential recurrence and ensure overall health and development.

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