SACROCOCCYGEAL TERATOMA - A CASE REPORT AND REVIEW OF LITERATURE

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ABSTRACT: Sacrococcygeal teratoma is a teratoma (kind of tumour) located at the base of coccyx (tail bone) and thought to be derived from embryonic germ cell layers ¹ ². They present mostly in infancy and are extremely rare in adults. Modern imaging techniques may be helpful to detect the extent of mass, but surgical excision is generally indicated at the time of detection. Most Sacrococcygeal teratomas are not likely to be malignant and prognosis tends to be good after resection. Here we are reporting a case of sacrococcygeal teratoma.

Keywords: Sacrococcygeal Teratoma, Management.

INTRODUCTION
Sacrococcygeal teratoma (SCT) is rare and happens in 1:35,000 to 40,000 live births. It is more common in girls than boys ratio of 3:1 to 4:1 have been reported ³ ⁴. The cause is unknown. This kind of tumor found to be made of different cells found during early development. Most Sacrococcygeal teratomas are found in neonates, infants and children below 4 yrs but reported in adult also. The tumour usually has both solid and cystic (fluid filled) parts. Some of the solid tumours have very large blood supply and can cause problem during pregnancy and the postoperative period. Like other teratomas Sacrococcygeal teratomas can grow very large even larger than the rest of baby. Large tumours can extend into pelvis and can affect other organs. The tumours that are found in new born period are usually benign but few can be malignant⁵⁶.

Sacrococcygeal teratomas can be classified by Altman⁵ based on the position of tumour, as follows:
Type 1- completely outside the body
Type 2- mostly outside the body with very small part in the pelvis
Type 3- some part outside the body with some part inside
Type 4- completely inside the body

In contrast to the newborn with SCT, fetus with SCT remains at high risk of perinatal complications and death. Fetuses with SCT detected antenatally have three times mortality rate compared to the fetuses with SCT diagnosed postnatally. Neonatal death may result from maternal obstetric complications as tumour rupture⁷, preterm labour or dystocia. The fetus is also at risk of high output cardiac failure⁸, placetomegally and hydrops with subsequent fetal demise secondary to the increased metabolic demands and vascular steal of a rapidly growing solid tumour⁹ ¹⁰. Modern technology using three dimensional (3D) sonography now allows the prenatal diagnosis of SCT even in the first trimester¹¹ ¹². Surgical resection remains the mainstay of management¹³ and after complete surgical excision recurrence is rare. This case is reported to highlight the clinical presentation and management of SCT in neonate.
CASE REPORT

23 year primigravida with 33 weeks gestation referred to our institution after sonographic finding of sacrococcygeal mass (teratomas) of size 9.5cm X 10cm for further management. That was her first antenatal visit. She was hemodynamically stable and not having any complains and all biochemical investigations(hemogram, complete blood count, renal function tests, kidney function tests, blood sugar, urine examination, PT/INR and ECG) were normal. Obstetric ultrasound showed 33 weeks live fetus with sacrococcygeal mass (of size 9.5 x 10 cm) arising from sacrum with adequate amniotic fluids and other parameters normal. After admission in OBGY ward on second day she went into labour. In view of large sacrococcygeal teratoma she underwent uneventful emergency cesarean section under spinal anaesthesia. Female baby delivered cautiously. Baby cried well after birth and with a large SCT 10 X 10 cms arising from sacral region with normal spine above (fig 1). Dilated veins above the swelling could be seen. (Fig 2). Baby weight was 2.5 kg and hemodynamically stable with good power in all four limbs. Child shifted to NICU for monitoring and in view of prematurity. X ray and MRI spine was done (fig 3) showing clear demarcation between tumour and spine. After 1 week baby posted for SCT excision. All biochemical investigations were within normal limit (Hb 13 gm%, sr creatinine 1.1mg/dl, BUN 29 mg/dl, Blood Sugar 100mg/dl, sr Bilirubin 1.2 mg/dl, SGOT 22 IU/dl, SGPT 23IU/dl, Sr Alkaline phosphatase 112 IU/dl, PT 14(15), INR 1, Blood group A positive, Sr electrolytes and urine examination normal) CXR was within normal limit.2D Echo showed tiny patent ductus arteriosus and mild LV dysfuction.USG abdomen was within normal limit and no bowel bladder involvement. From radiological investigations it was arising from coccyx, type 1 SCT with mass effect on rectum.

Baby kept NBM (nil by mouth) for six hours prior to operation for this period baby received maintenance fluid (5% dextrose in 0.45% normal saline) 10 ml/hr. Same intravenous fluid continued perioperatively. Premedicated with 0.01 mg glycopyrrolate and fentanyl 5 microgram Preoxygenation done for 3 mins. Before induction baby placed on soft pillow with tumour supported with soft cotton rolls beneath and around. Induction done with sevoflurane, after induction baby intubated with plain portex endotracheal tube no.2.5. Anaesthesia maintained with oxygen and nitrous oxide 50:50 and sevoflurane 1-2%. For muscle relaxation atracurium 1.25 mg iv given as loading dose followed by two top ups of 0.25mg. Baby kept warm by wrapping limbs and head in cotton rolls. ECG, PR and Spo2 was monitored intraoperatively. Surgical resection of tumour including coccygectomy was done. At the end of surgery spontaneous respiratory attempts were good, so baby reversed with neostigmine 0.125 mg and Glycopyrrolate 0.2 mg. After regaining adequate muscle tone, cough and gag reflex, baby extubated. Intraoperative blood loss included blood loss at operative site as well in excised tumour (large dilated veins) and was around 70 ml. Blood loss replaced with 70 ml of packed cell volume. 3ml of calcium gluconate was given by iv infusion (as ionic calcium level decreases after blood transfusion due to binding of citrate with calcium). Intraoperative urine output was maintained and around 1ml/kg/hour. Postoperatively baby shifted to NICU for further monitoring and management. In NICU baby received oxygen with hood and dobutamine infusion 2.5microgram/kg/min (in view of low volume pulse and mild LV dysfunction on ECHO) for 24 hours. For postoperative pain management tramadol 5mg 12 hourly given. Anaesthetic challenges were mainly prematurity and blood loss. Histological examination of excised tissue was consistent with cystic and solid parts with no evidence of malignancy. The baby had an uneventful postoperative recovery and discharged home after 15 days.
Photo 1: showing sacrococcygeal teratoma almost equal to baby size

Photo 2: showing lateral aspect of baby and SCT with dilated vein

Photo 3: X ray of baby involving whole body and SCT showing no bony involvement

DISCUSSION
The newborn with Sacrococcygeal teratomas (SCT) has an excellent prognosis depending on the time of diagnosis, malignant potential of the tumour and the ease of surgical excision. Early prenatal diagnosis is
possible, but our patient did not attended regular antenatal check up. She went for ANC visit at 33 weeks only and USG done in same visit revealed fetus with SCT. SCT could be diagnosed from second trimester of pregnancy when there is polyhydraminos and/or uterus larger than the gestational age. Prenatal diagnosis is of significance, since early prenatal presentation is associated with high fetal morbidity and mortality while presentation after 30 weeks is a relatively good prognostic indicator for fetal survival\textsuperscript{4}.

Monitoring for fetal distress during pregnancy is very important. Some large tumours have a very high blood flow that causes a shift in blood flow away from the baby towards the tumour. As it grows it can cause the baby to become sick and hydropic. This means the heart begins to fail and the baby becomes swollen. Other possible complications include bleeding inside the tumour, development of excess amniotic fluid and preterm labour. Progressive hydrops can be associated with a swollen and sick placenta. There is a rare condition called ‘Mirror syndrome’. Where the mother mirrors the baby’s sickness\textsuperscript{5}. If this occurs baby should be delivered.

Early diagnosis may predicate delivery by cesarean section in centres with good neonatal facilities where early surgical treatment can be offered to the baby\textsuperscript{1,10,17}. That’s what happened with our case due to prenatal diagnosis baby delivered safely with cesarean section and also surgical excision was done at earliest. Similarly fetal surgical procedures\textsuperscript{18} could be undertaken when the diagnosis is made early in the pregnancy. Well planned surgical excision was undertaken in our patient, care also taken to ensure inclusion of the coccyx in the excision material in order to forstall possible recurrence. Apart from age at diagnosis, treatment and the extent of resection, the prognosis is also determined by the histological type and stage at the time of resection and not the size of tumour. Complete excision including coccygectomy as done in our case, is the primary therapy for all SCT and it is adequate\textsuperscript{2,15} if the tumour is benign. Chemotherapy and radiotherapy are however indicated in malignant cases. In case of malignancy recurrence is as high as 37\% has been reported if coccyx is not removed in the primary surgery.

Follow up in patients with SCT is necessary especially during the first three years of treatment\textsuperscript{19,20}. When recurrence is likely. Extensive surgery in the pelvis and perineal region may involve disruption of nerves and muscles which supply urinary/anorectal sphincters and provide maximum support in normal working respectively. Alpha fetoprotein well known marker of teratomas, is valuable in differentiating between mature and malignant teratomas at presentation and during follow up of patients\textsuperscript{21}. It may be utilized to detect early occurrence of malignancy. This is not only appropriate during first three postoperative years when recurrence is likely, but on long term basis as significant number of them suffer from deficient anorectal function and diminished quality of life\textsuperscript{20,21,22}. Sacrococcygeal teratoma is a rare tumour that may be benign or malignant. Complete excision is the primary therapy, and is adequate if tumour is benign. Chemotherapy and radiotherapy are however indicated in malignant cases and in recurrence after previous excision.

REFERENCES