Case Report

Sacrococcygeal teratoma: A case report

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ABSTRACT

Sacrococcygeal teratomas (SCTs) are the most common extragonadal germ cell tumors, comprised different types of tissues that come from at least two of three germ cell layers. Depending on the tissues that are included, they are divided into mature, immature, and malignant. The incidence of SCT in infants and children is 1 in 35,000–40,000 live births. We are reporting a case of type I SCT. The patient was gravida 3, para 1, and abortion 1, which was diagnosed during antenatal ultrasound examination at 22 weeks of gestation and the termination was done after counseling the parents. A female fetus with a tumor in the sacrococcygeal region, weighing 800 g was delivered. The baby was sent to the department of anatomy. SCTs develop at the base of the coccyx and are thought to be derived from Henson's node a rounded and elevated area at the cranial end of the primitive streak. This primitive streak consists of totipotent cells, which are able to transform into any type of cells.

Keywords: Extragonadal germ cell tumors, Henson's node, Primitive streak, Sacrococcygeal teratoma, Totipotent cells

Sacrococcygeal teratomas (SCT) are the most common extragonadal germ cell tumors (GCT) in infants and young children. It is common in female babies with a ratio which is 1:3 to 1:4. GCT are neoplasms that develop from primordial germ cells of the human embryo. SCT represents the most common benign GCT in children accounting for up to 70% of all GCT. Worldwide, the incidence varies from 1 in 35,000 to 40,000 live births. During the neonatal period, they present with visible external sacral mass, but some patients with intra-pelvic tumors may present later [1,2]. SCTs are comprised 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, and malignant teratomas [2]. In contrast to the newborn, the fetus with SCT remains at a high risk of perinatal complications and death. Neonatal death may result from maternal obstetric complications of tumor rupture, preterm labor, and dystocia. The fetus is also at the risk of high output cardiac failure, placentomegaly, and hydrops with subsequent fetal demise secondary to metabolic demands and a vascular steal of rapidly growing solid tumor [3].

The present case was diagnosed prenatally as type I SCT at 22 weeks of gestation. It is reported due to the rarity of the anomaly.

Access this article online	
Received - 22 December 2021 Initial Review - 06 January 2022 Accepted - 27 April 2022	Quick Response code
DOI: 10.32677/ijcr.v8i5.3200	

CASE REPORT

A 22-year-old woman was admitted at 22 weeks gestation to our hospital. She was gravida 3 and para 1, 3 years living male baby and 1 abortion at 10 weeks. Her LMP was 11/01/21. There was no history of consanguineous marriage and no family history of any anomalous babies. She was advised to take folic acid 5 mg daily from 6 weeks onward.

On examination, her blood pressure was 120/70 mm Hg. The patient was mildly anemic and there was no edema of the feet. Clinical examination of the cardiovascular system and the respiratory system was normal. Per abdomen examination showed uterus of 24–26 weeks relaxed.

Urine analysis was normal, hemoglobin was 10.3 g%. An ultrasound scan done at 12 weeks showed a single live intrauterine pregnancy with no detected anomalies. The second-trimester scan done at 22 weeks of gestation showed features of SCT. A fairly ill-defined mixed echogenic solid cystic mass lesion was seen in the sacrococcygeal region. It was measuring 6.9 × 6.3 × 7.9 cm with few internal hypoechoic foci, likely calcifications. No evidence of intra-pelvic extension of the mass lesion was seen (Fig. 1). On color Doppler, the lesion showed significant internal vascularity, likely SCT type I with minimal fetal ascites and polyhydramnios. The biparietal diameter was 5.18 cm, head circumference was 18.71 cm, abdominal circumference (AC) was 16.63 cm, femur length (FL) was 3.41 cm, fetal heart rate was 161/min, FL/AC

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was 1.31, AC was 1.31, and estimated fetal weight was 409 g. All parameters correspond to 21 weeks. Amniotic fluid index was 21 cm, cord 3 vessel, placenta posterior, and grade II. Cranium, lateral ventricles, face, and spine appear normal.

The couple was counseled for the termination of pregnancy. After induction, she expelled a female fetus weighing 800 g including the placenta (Fig. 2). On examination, a tumor measuring 9 × 10 × 7 cm was seen in the sacrococcygeal region partly covered with a membrane.

DISCUSSION

SCTs are common tumors diagnosed in the neonatal period. Teratoma in children originates most commonly in the sacrococcygeal region but can be seen in other sites such as gonads and the retroperitoneal region. Rarely, it is seen in the brain and liver. The incidence of malignancy in the neonatal period is 10% against 100% at the age of 3 years [1]. Meena *et al*. [1] reported a retrospective analysis of neonates, infants, and children with SCT. During the study period of 2 years and 8 months (Jan 2014–Oct 2016), a total of 28 cases were evaluated, of which 21 were female and 7 were male (F: M 3:1). Nine neonates and 13 infants with externally visible mass were diagnosed as type I and type II, respectively, and six presented after 1 year. The present case was also a female baby and diagnosed with type I, which was similar to the study reported by Meena *et al*. Boro *et al*. [2] reported a case G2 P1 diagnosed with type I SCT at 20.3 weeks by magnetic resonance imaging scan. The patient went into preterm labor at 35.1 weeks, a lower segment cesarean section (LSCS) was done and a male baby was delivered. After 36 h, embolization of the middle sacral artery and excision of the tumor was done and the baby went home alright.

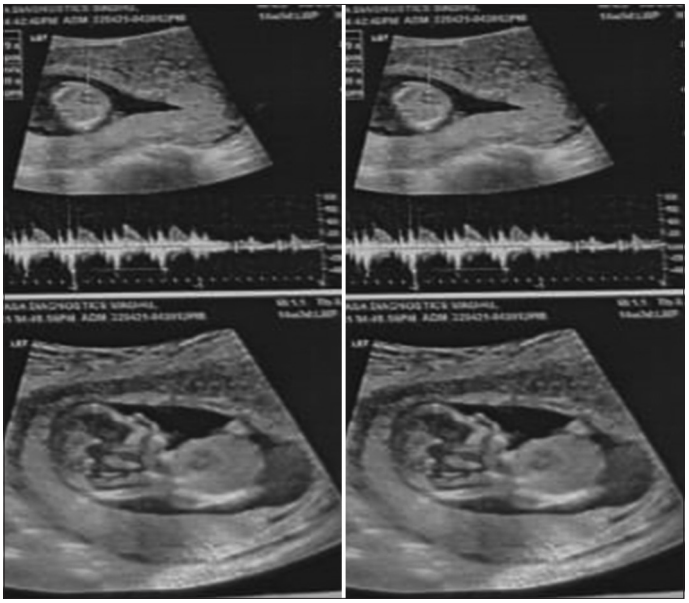

Figure 1: Ultrasound showing sacrococcygeal teratoma at 22 weeks

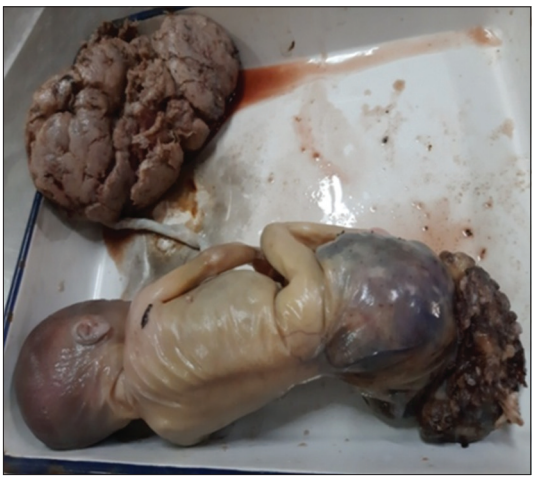

Figure 2: Photograph of baby with huge sacrococcygeal teratoma along with umbilical cord and placenta

According to the American Academy of Pediatrics Surgical Section, SCT is classified into four types in 1973. Altman classified SCT into four types based on the external components and intra-pelvic/intra-abdominal extension of the tumor [4]. Type I is predominantly external, type II presents externally with significant intra-pelvic extension, type III is apparent externally but predominantly pelvic mass extending into the abdomen, and type IV is presacral with no external presentation [5]. SCT seen at birth is usually type I and II (87%), rarely type III can be seen, and type IV is typically seen later in life as there is no external component [4].

According to the previous literature, diagnosis of SCT is made usually at 20–24 weeks of gestation [2]. A case report stated that a fetus can be diagnosed as having SCT as early as 12+3 weeks using 3D Ultrasound examination [6].

According to the embryological basis, SCTs develop at the base of the coccyx and are thought to be derived from Henson's node (Primitive streak), a rounded and elevated area at the cranial end of the primitive streak. The primitive streak is a longitudinal ridge of ectodermal cells at the caudal end of the bilaminar germ disk. This structure appears at the beginning of the 3rd week and is formed by the proliferation of the ectodermal cells which move toward the midline of the embryo. This primitive streak consists of totipotent cells, which are able to transform into any type of cells. After the formation of the intraembryonic mesoderm, the primitive streak completely disappears by the end of the 4th week or it becomes an insignificant structure in the sacrococcygeal region of the embryo. If totipotent cells remain after the 4th week, these cells give rise to SCT. The degree and type of differentiation of these cells decide the specific type of tumor. At birth, the majority are benign [7].

Rattan and Singh [8] reported a study of retrospective analysis of neonates with SCT operated from 1997 to 2016. Among the 44 enrolled, the prenatal diagnosis of SCT was available in 25% of pregnancies. All babies except one were born vaginally. Associated anomalies were seen in 20%. Type I was 27.3%, type II was 54.5%, type III was 13.6%, and type IV was 4.5%. Morphologically, 77% of neonates had cystic lesions. All were managed successfully by local excision. Legbo *et al*. [3] reported a case of 10-day-old male baby with mature SCT. A mass consisting of a fully developed lower limb was attached to the sacrococcygeal region. The lower limb was a bag of skin like an empty scrotal sac and more medially was a roll of three teeth. Bagle *et al*. [9] reported a case of a female baby

weighing 2.5 kg with SCT delivered by LSCS at 33 weeks and the excision was done after a week. Sinha *et al*. [4] reported a study of ten cases with SCT who underwent successful surgical excision at the mean age of the 9th postnatal day. The present case was diagnosed as type I SCT at 21 weeks of gestation and the ultrasound report showed signs of calcification in the mixed echogenic mass and the fetus had signs of ascites, so termination was decided.

There is a high risk of perinatal complications and death which may result from tumor rupture, preterm labor, and dystocia. The fetus is also at risk of high output cardiac failure and hydrops with subsequent fetal demise secondary to metabolic demands and vascular complications.

CONCLUSION

SCT is a rare congenital malformation and prenatal diagnosis is important to plan for delivery and planning surgical excision during the postnatal period.

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Funding: None; Conflicts of Interest: None Stated.

How to cite this article: Menasinkai SB, Chiniwar MA. Sacrococcygeal teratoma: A case report. Indian J Case Reports. 2022;8(5):125-127.