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Case Report

Sacrococcygeal Teratoma: A Case Report

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ABSTRACT

Background: Neonatal tumors are usually prenatally diagnosed or within the first 30 days of life. The true incidence of neonatal tumors is unknown as a great number of pregnancies with a prenatally diagnosed mass result in stillbirth or miscarriage. Most solid neonatal tumors are benign whereas less than 50% of neonatal neoplasms are malignant; however, some tumors with malignant patterns may histologically show benign behaviors. The incidence of malignant tumors is 1 in every 12,500-27,500 live births, accounting for 2% of all childhood cancers. Teratomas are the most common perinatal neoplasms, accounting for 25-33% of the cases. Around two-thirds of all sacrococcygeal teratomas (SCTs) are reported in the neonatal period but with a small risk of malignancy.

Case report: Herein, we reported a case of SCT in a newborn leading to a complicated cesarean delivery.

Conclusion: In general, teratomas are embryonic typically benign tumors arising from germ cells. They usually consist of various tissues originating from two or more embryonic layers. These tumors are believed to be the most common neonatal germ cell tumors. The sacrococcygeal region is the most common site for extragonadal teratomas formation.

Keywords: Neonatal tumors, Neoplasm, Teratoma

Introduction

Sacrococcygeal teratoma (SCT) with the incidence of 1:25000 live births is the most common type of tumor presenting at birth (1, 2). The tumor is mostly seen in females with a female:male ratio of about 3-4:1 (3, 4). According to the American Academy of Pediatrics Surgical Section, SCT is classified into four types regarding its location (Figure 1). Type I is an external tumor with minimal presacral components whereas type II has significant extensions into the pelvis; these are the most common types presenting at birth. Type III, which is rare at birth, is apparently external but has significant extension into the abdomen. Type IV, which typically presents later in life, is a presacral mass without external manifestation (1, 5, 6).

If SCT is antenatally diagnosed in a fetus, it can lead to a three-time higher risk of mortality, compared to those diagnosed after birth (4). Fetal complications associated with SCT include high-output cardiac failure resulting in nonimmune fetal hydrops, internal or external hemorrhage, as well as dystocia and preterm labor due to polyhydramnios (2, 4, 7). If not antenatally

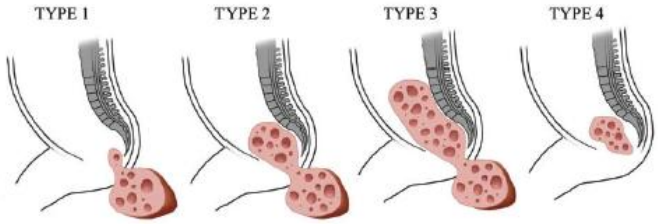

Figure 1. Morphological classification of sacrococcygeal teratomas according to the American Academy of Pediatrics Surgical Section Survey

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diagnosed, SCT may also result in traumatic delivery (2). Nevertheless, SCT and its complications are usually diagnosed in the second trimester through ultrasound (8).

Case report

A newborn was visited by a pediatric resident in the operation room upon the urgent request of an obstetrics resident. The newborn was a girl with 37 weeks of gestation delivered through cesarean section. Her 5- and 10-minute Apgar scores were 9 and 10, respectively. She was born on May 6th 2019 in Imam Reza Hospital in Mashhad, Iran. Her father and her mother were 35 and 27 years old and non-related. Her mother was diagnosed to have hypothyroidism and hypertension during pregnancy, treated with Levothyroxine and Methyldopa, respectively. The parents' medical history had nothing remarkable to disclose. The mother had not undergone any perinatal ultrasounds so the exact time of tumor formation was unknown.

When she was admitted for delivery, an ultrasound was performed revealing a cystic focus with midline septa at the sacrococcygeal region suggestive of a meningocele. The delivery was performed resulting in a hemodynamically stable newborn with a normal respiratory pattern and pinkish skin color. No heart murmur was detected, and the abdomen appeared normal on physical examination. However, a mass of about 20 × 10 cm in size was noticed at the sacrococcygeal area (Figure 2), and the perforated anus had an unusual position at the upper right side of the mass. The left thigh seemed immobile with a tender swelling but with no discoloration.

The newborn was immediately transferred to the neonatal intensive care unit (NICU) for further evaluation. Feeding initiated and was tolerated quite well. The thoracoabdominal and lower limb X-ray was taken revealing a mass with fine calcifications on the right side, along with fracture of the left femoral shaft (Figure 3). Ultrasound of the mass and abdomen was normal except for a massive cystic mass, including septa in the sacrococcygeal region suggestive of type I teratoma. The orthopedic consult was requested that suggested the application of a long leg splint since casting was impossible regarding the tumor's position. A pediatric surgery consult was also conducted, and the newborn was transferred to the pediatric surgery ward for the complete resection of the tumor and coccyx.

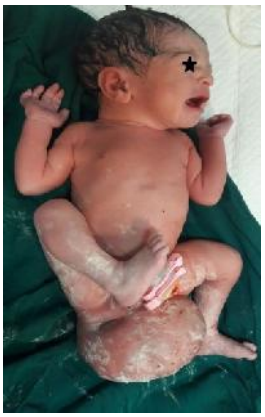

Figure 2. The newborn (with 37 weeks of gestation) with a sacrococcygeal teratoma

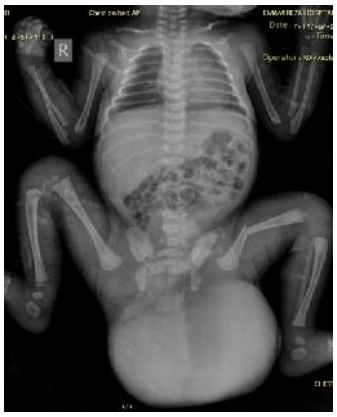

Figure 3. Anteroposterior thoracoabdominal and lower limbs X-ray; observation of large teratoma and fracture of the femoral shaft

Discussion

The SCTs are extragonadal neoplasms composed of different types of tissue and various sizes located in the presacral region arising from two or three germ cell layers (9). Our patient was a girl that complies with the female predominance reported in such cases in the literature (3, 4). In a study by Altman et al., SCTs were classified into three groups based on their size and diameter as small (2-5 cm), moderate (5-10 cm), and large (>10 cm) (7); accordingly, our patient had a large tumor. On ultrasound, SCT was reported as type I that is the most common type in neonates and has a low risk of developing malignancy (1, 5).

The most common site for teratomas is the sacrococcygeal region but they can also be located in the mediastinum, testes, retroperitoneum, brain, head and neck, vagina, stomach, and pineal region (1, 10). One of the most important differential diagnoses of SCT, especially type I, is myelomeningocele (8) suggested by the ultrasound performed before delivery. However,

the ultrasound conducted in the NICU diagnosed the mass as an SCT.

The larger the sacrococcygeal tumors, the more complex the delivery might be as larger tumors are associated with the increased risk of rupture, hemorrhage, and dystocia during delivery (1, 9). Teratomas developed in the early fetal period entail a high risk of morbidity and mortality. In contrast, teratomas presenting after 30 weeks of gestation are associated with a relatively good prognosis (4).

In our case, an ultrasound was performed at admission in which the newborn's SCT was diagnosed. Since she had not undergone any perinatal ultrasound, the exact time of tumor formation remained unknown. Although the obstetrician planned a cesarean delivery regarding the large SCT to avoid probable complications, the baby suffered from left femur fracture while being pulled out.

Complete surgical resection of the tumor and coccyx still remains the primary treatment for SCT; if they are left untreated, there would be a high risk of recurrence (1, 2, 7, 11). Therefore, our patient was referred to the pediatric surgery ward for the complete excision of the tumor and coccyx. She also received the optimal treatment for her femoral fracture, including traction-suspension of both lower extremities (2). Due to the position of the tumor, it was impossible to immobilize the lower limbs in spica casts, so a long leg splint was used to temporarily stabilize the injury, and the standard treatment of the fracture was postponed to after the main surgery.

Conclusion

The SCTs are the most common tumors presenting at birth. They occasionally are accompanied by complications, such as rupture, bleeding, hip dislocation or limb fractures. Our case was a neonate born with a mass in the sacrococcygeal region diagnosed as a teratoma. The large mass led to left femoral fracture during delivery. The mainstay of treatment remains surgery performed for our patient. This case report once again emphasized the possible association of SCT with birth injuries and complications and the importance of seeking the safest route of delivery to minimize these undesired events.

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