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**Case Report**

# Antenatal teratoma sacrococcygeal in a three month old children in Indonesia: a rare case report

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## ABSTRACT

Sacrococcygeal teratoma is a type of germinal cell tumor in children, usually observed in the prenatal or antenatal phases. In either phase, sacrococcygeal teratoma may cause complications that lead to increased risk of morbidity and mortality. A proper screening and management for sacrococcygeal in children are mandatory for the best outcome. Currently, surgical resection is the most promising treatment to manage antenatal sacrococcygeal teratoma in children. Three-month-old female children were brought to the hospital with a growing 8.4 cm mass since birth in her groin. The mass was painless and soft in consistency. Computed tomography (CT) scan revealed a dominant solid lesion accompanied by multiloculated fluid density arising from right gluteal muscle. The patient planned for surgical excision. Hyperkalemia was observed prior to the operation; hence an appropriate correction was needed before surgery. Germ cell tumors, though uncommon, may develop beyond the gonadal region, such as in the brain, neck, mediastinum, and retroperitoneum. In neonates and fetuses, these tumors frequently originate in the sacrococcygeal area, possibly resulting from the migration of primordial cells and the proliferation of totipotent stem cells in that region. Optimal disease burden outcomes for sacrococcygeal teratoma are often achieved through surgical resection. Sacrococcygeal teratoma, although rare, might possess a mortal threat to patient, hence proper diagnosis and treatment must be done to ensure the best clinical outcome.

**Keywords:** Antenatal sacrococcygeal teratoma, Children, Germ cell tumor

## INTRODUCTION

Sacrococcygeal teratoma is a type of germinal cell tumor in children, especially in prenatal to antenatal phases.[1] Germinal cell tumors are considered uncommon, with an incidence of 2.4 cases per million births per year or less than 3% of incidence of malignancies in pediatric populations overall.[2,3]

Ultrasound is commonly used to screen fetal sacrococcygeal teratoma.[4] In some cases, magnetic resonance imaging (MRI) can help supplement a more precise view of sacrococcygeal teratoma and its implications to nearby organs.[4] In cases of prenatal discovery of sacrococcygeal teratoma, complications can and may increase the risk of intrauterine fetal death up to three-fold (compared to cases of sacrococcygeal teratoma discovered antenatally).[5] This can be caused by fluid retention in the tumor which can reduce fetal cardiac output.[5,6] This phenomenon is known as hydrops fetalis and is main indication for fetal surgery.[5,6] Techniques currently known to treat cases of sacrococcygeal teratoma in fetuses include endoscopic laser ablation and intra-tumor radiofrequency ablation.[4]

Sacrococcygeal teratoma can be diagnosed during the antenatal period. In instances where sacrococcygeal teratoma is detected before birth, the optimal treatment approach involves the complete excision of the teratoma.[5,7] Nevertheless, it's important to note that recurrences of sacrococcygeal teratoma have been documented in 10% to 20% of cases even after the thorough removal of the teratoma.[7]

## CASE REPORT

A 3-month-old girl (weight 7.5 kg and body length 60 cm) came to the hospital with complaints of a lump in the groin area. According to the patient's family, the lump was felt to have been present since birth and grew bigger over time. A mass measuring 8.4 cm with a soft and painless consistency was observed during the initial exam. Other complaints such as fever, wounds or signs of inflammation in the mass area, decreased appetite, or lumps in other body areas were denied. Vital signs and other physical examinations were within normal limits.

CT scan revealed a dominant solid lesion accompanied by multiloculated fluid density accompanied by calcified components in the soft tissue of the right gluteal region which was clearly bordered by the right gluteus maximus muscle. The lesion is not related to a defect in the sacral spinal canal, does not infiltrate the abdominal cavity or pelvis. The lesion is attached to the right side of the anal dimple. The CT scan suggests an immature teratoma. There was also multiple reactive lymphadenopathies in the inguinal and axillary regions bilaterally. Laboratory examination (Table 1) showed hyperkalemia (6.5 mmol/l). The patient was planned to undergo an elective procedure for excision of the mass after correction of hyperkalemia.

**Table 1: Preoperative laboratory testing.**

| Parameters | 22/9 | 23/9 | 24/9 | 25/9 | 26/9 | 27/9 | Units |
|---|---|---|---|---|---|---|---|
| **Hemoglobin** | 11.4 | | | | | | g/dl |
| **Hematocrit** | 33 | | | | | | % |
| **Leukocyte** | 8.1 | | | | | | $10^3$/µl |
| **Platelet** | 326 | | | | | | $10^3$/µl |
| **Erythrocyte** | 3.83 | | | | | | $10^6$/µl |
| **MCV** | 86.4 | | | | | | /µM |
| **MCHC** | 34.4 | | | | | | pg |
| **MCH** | 29.7 | | | | | | g/dl |
| **Neutrophil** | 30.30 | | | | | | % |
| **GDS** | 96 | | | | | | mg/dl |
| **PT** | 11.4 | | | | | | s |
| **APTT** | 37.5 | | | | | | s |
| **INR** | 0.81 | | | | | | |
| **Urea** | 14 | 9 | | | | | mg/dl |
| **Creatinine** | | 0.2 | | | | | mg/dl |
| **Na** | 135 | 137 | 131 | | | 139 | mmol/l |
| **K** | 6.5 | 6.1 | 5.4 | 5.1 | 5.4 | 4.4 | mmol/l |
| **Cl** | 105 | 108 | 99 | | | 104 | mmol/l |
| **Mg** | | 0.53 | 0.5 | | | | mmol/l |
| **Ca** | | | 1.32 | | | | mmol/l |

In the course of addressing hyperkalemia, the patient underwent treatment administered by the pediatrician. Intravenous furosemide (0.5 mg/kg/8 hours) and insulin (0.1 IU/kg) which were given simultaneously with a fluid cycle of 20 cc of 5% dextrose was administered in order to correct hyperkalemia. The patient was also given a cycle of calcium gluconate (50 mg/kg) diluted in 40 cc of 5% dextrose. After the hyperkalemia was managed, the patient went for surgery.

The surgery commenced after the patient was under general anesthesia. The patient was positioned supine and continued with draping and disinfection of the surgical area. The first incision began directly above the tumor and advanced through its base. Tumor was visualized and measured approximately 8×4×3 cm. Tumor was excised by separating affected and unaffected area whilst suctioning excess bleeding. Wound closure was done layer by layer from inside out.

After 3 hours of surgery, the patient was transferred from the operating room to the pediatric intensive care unit (PICU) under sedation and a 3.5 size 10 cm deep endotracheal tube (ETT) was installed. The patient is reconnected to the ventilator in assist control/pressure control mode. Postoperative laboratory examinations were also carried out (Table 2).

After three days in PICU, the patient was transferred to the high care unit (HCU). Proper wound care was also

done during the recovery period in the HCU. During treatment in the ICU and HCU, packed red cells (PRC) transfusions are also carried out to correct anemia. Fluid was also given by administration of intravenous 5% dextrose infusion, paracetamol (15 mg/kg/8 hours), ampicillin (50 mg/kg/6 hours), and metamizole (15 mg/kg/8 hours). Oral intake is started three days after the surgery with a daily fluid requirement of 750 cc per day.

The baby was discharged at 7 days after operation with normal results of the scans. Follow-up was performed and no neurological deficits have appeared.

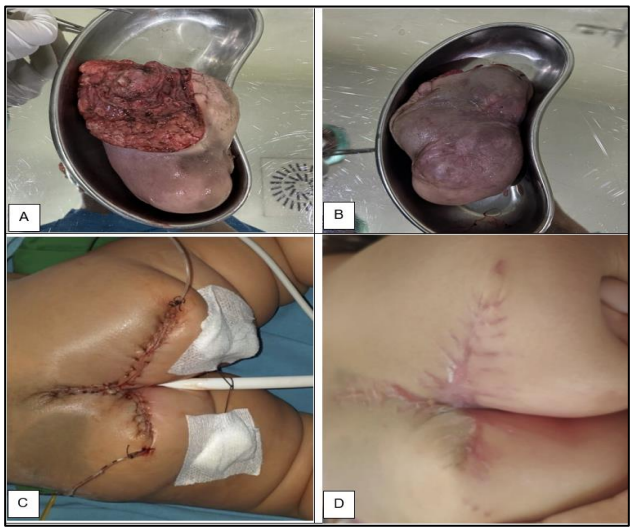


**Figure 1 (A-D): Post-excision mass appearance; post-excision surgical wound and post suture removal.**

**Table 2: Post operative laboratory testing.**

| Parameters | 29/9 | g/dl |
|---|---|---|
| **Hemoglobin** | 8.6 | % |
| **Hematocrit** | 26 | 103/µl |
| **Leukocyte** | 21.9 | 103/µl |
| **Platelet** | 462 | 106/µl |
| **Erythrocyte** | 3.01 | /µM |
| **Neutrophil** | 82.2 | mg/dl |
| **Blood gas analysis** | | |
| pH | 7.4 | |
| Base excess | 0.6 | |
| $PCO_2$ | 39.3 | |
| $PO_2$ | 121.8 | |
| Hematocrit | 30% | |
| $HCO_3^-$ | 25.8 | |
| Total $CO_2$ | 26.6 | |

## DISCUSSION

The majority of germ cell tumors originate from cells and develop in the gonads. In rare cases, germ cell tumors can also grow outside the gonad area such as in the brain, neck, mediastinum and retroperitoneum.[1] In the case of neonates and fetuses germ cell tumors most often arise from the sacrococcygeal area.[1] Although sacrococcygeal teratoma is considered most common in the antenatal and prenatal phases, its general incidence is considered rare (1:20,000-40,000 live births) and is predominantly female.[1,4]

There are two theories that explain why teratomas can grow in the sacrococcygeal region. The first theory relates to the process of migration of extragonadal primordial germ cells from the yolk sac through the hindgut where sacrococcygeal teratoma can grow.[1,4,6] The second theory explains that embryonic stem cells can differentiate into teratomas in the sacrococcygeal region. In the sacrococcygeal region there are remaining totipotent stem cells from Hensen's node which can differentiate into teratoma.[1]

In one third of cases, patients with sacrococcygeal teratoma may suffer from other congenital abnormalities such as hydronephrosis.[1] In addition, in cases where the sacrococcygeal teratoma is large, the risk of the neonate suffering from consumptive coagulopathy, thrombo-cytopenia, disseminated intravascular coagulation, and bleeding is increased.[1] In fetal sacrococcygeal teratoma, a growing mass can affect fetal haemodynamic by reducing venous return and decreasing cardiac output due to hydrops fetalis.[5]

Currently, surgical resection is the main strategy to manage sacrococcygeal teratoma. The prognosis and postoperative outcome of the resection were assessed as good.[1] Recurrence is reported in 10%-20% of cases, with one of the risk factors for recurrence being incomplete recurrence. Another risk factor for recurrence is the malignant subtype of teratoma which is more frequently found in adult.[1] To prevent incomplete resection, coccyx bone is often removed to avoid the potential growth of residual tumor cells.[1]

In this case was reported a sacrococcygeal teratoma present in a three-month-old female baby that arises since birth. No functional or anatomical abnormalities were observed related to the growing mass. This case is considered unique in our center considering the history of illness, age, gender, and the nature of the mass itself. We carried out an excision of the mass and the surgery was successful in order to remove the mass. Postoperatively the patient also doing well, with proper wound healing was observed weeks after surgery.

## CONCLUSION

Antenatal sacrococcygeal teratoma, although rare, can present a significant and potentially mortal threat to the patient. This congenital tumor, originating at the base of the spine, exhibits a diverse range of tissues, including those derived from all three germ layers. Due to its intricate characteristics, an accurate and prompt diagnosis is crucial for establishing an effective surgical treatment strategy if needed. Surgeons must employ a

multidisciplinary approach, involving specialized imaging techniques, such as ultrasound and magnetic resonance imaging, to precisely characterize the tumor and assess its potential impact on surrounding structures.

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