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(CASE REPORT)

Defying the Odds: Successful surgical management of a rare intrapelvic sacrococcygeal teratoma in Eastern Uganda

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Abstract

Sacrococcygeal teratomas (SCTs) are the most common congenital tumours in neonates, typically presenting as external masses at birth. We present a rare case of a predominantly intrapelvic SCT in a one-month-old female infant from rural eastern Uganda. The child was referred with a progressively enlarging sacrococcygeal mass that caused anal deviation and difficulty in defecation. Clinical evaluation and imaging suggested a presacral mass without distant spread. Surgical exploration via a posterior sagittal approach revealed a solid intrapelvic tumour tightly adherent to the posterior rectal wall and coccyx. Complete excision of the mass, including coccygectomy, was successfully performed without injury to adjacent structures. Histopathology confirmed a mature sacrococcygeal teratoma. Postoperative recovery was uneventful, and follow-up showed no signs of recurrence or functional impairment. This case underscores the importance of early recognition of SCTs, even in atypical presentations without external components. It also reinforces the surgical principle that complete resection with coccygectomy is essential to reduce the risk of recurrence. Despite the challenges of a low-resource rural setting, this case illustrates that successful management of complex congenital tumours is achievable with timely intervention and appropriate surgical planning.

Keywords: Sacrococcygeal teratoma; Coccygectomy; Intrapelvic tumour; Neonatal surgery; Anorectal distortion; Paediatric surgical oncology

1. Introduction

Sacrococcygeal teratoma (SCT) is a congenital germ cell tumour that arises from pluripotent cells in the sacrococcygeal region and contains elements derived from all three germ layers: ectoderm, mesoderm, and endoderm [1,2]. It represents the most common extragonadal germ cell tumour in neonates, with an estimated incidence of 1 in 35,000 to 40,000 live births and a marked female predominance (female-to-male ratio of approximately 4:1) [3]. SCTs are typically diagnosed at birth or antenatally, often presenting as large external masses. However, intrapelvic or presacral variants especially those without prominent external components are less common and pose significant diagnostic and surgical challenges.

Although SCTs are well-documented in high-resource settings, there is a dearth of literature from sub-Saharan Africa, particularly regarding intrapelvic presentations and their management outcomes. In low-resource environments, delayed diagnosis, limited access to imaging, and constrained surgical capacity further complicate care. Despite these barriers, timely surgical intervention remains the cornerstone of effective treatment, with complete excision including coccygectomy being critical to prevent recurrence.

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We report a rare case of a predominantly intrapelvic mature SCT in a neonate from rural eastern Uganda, managed successfully through complete surgical resection in a resource-limited setting, highlighting both the clinical and logistical challenges encountered [4].

2. Case Presentation

A one-month-old female infant was referred to our surgical unit with a progressively enlarging mass over the sacrococcygeal and perineal region, first noted at birth. She was born at 37 weeks and 2 days of gestation via spontaneous vaginal delivery at a peripheral Health Centre IV to a 17-year-old primigravida. The mother had attended antenatal care only three times during the pregnancy and commenced routine supplementation with folic acid and iron in the second trimester. No antenatal ultrasound was performed. She lived with her husband and reported no complications during pregnancy.

Labor began spontaneously at home around midnight. She was transported by motorcycle (boda-boda) and arrived at the health facility at 1:00 am. A normal labour course followed, and at 4:00 am, she delivered a live female infant who cried immediately after birth. The newborn had APGAR scores of 8 and 10 at 1 and 5 minutes, respectively, and weighed 3.01 kg.

A midwife conducting the initial neonatal examination noted a swelling at the lower back, which she reported to the mother. The attending clinicians subsequently recommended an urgent referral to Mbale Regional Referral Hospital. However, the mother requested to return home first to collect necessary items before traveling to the referral center, which is approximately 60.7 km away.

The infant was admitted to the neonatal unit on the third day of life, where she remained for one week before being discharged with instructions to return for surgical evaluation at one month of age. At follow-up, the mass had significantly increased in size, displacing the anal opening anteriorly. Although the child was able to pass stool, the mother reported abnormally frequent defecation. The infant was otherwise feeding well and had no signs of systemic illness.

On physical examination, a firm, non-pulsatile mass was noted in the sacrococcygeal region, extending into the gluteal cleft and inferiorly displacing the cleft. The anus was displaced anteriorly and inferiorly (figure 1). No signs of infection or overlying skin ulceration were observed. Rectal examination was limited due to the distortion but revealed no obvious mucosal lesions.

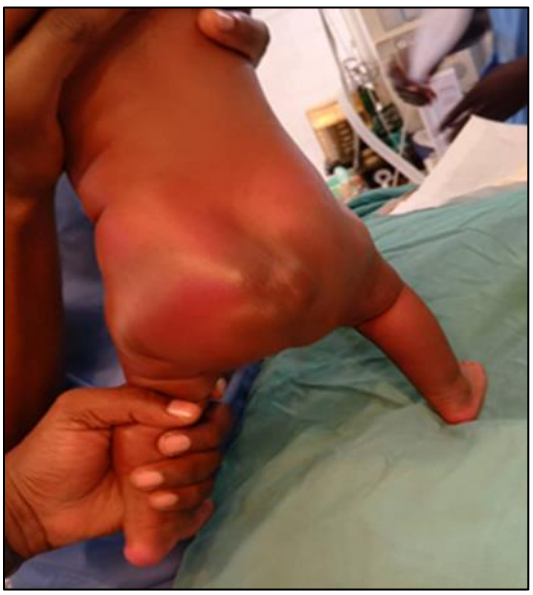

Figure 1 Physical examination

Ultrasound of the lumbosacral region revealed a well-defined, echogenic complex mass located in the sacrococcygeal area. Sonographic features demonstrated a heterogeneous lesion with both solid and cystic components, as well as peripheral vascularity on colour Doppler. The mass measured approximately 62.7 × 65.0 × 56.3 mm, with an estimated

volume of 120 cm^3, and was predominantly solid. It extended into the pelvic cavity and appeared to be adherent to the sacrum and coccyx, causing anterior displacement of the rectum. The vertebral column appeared normal, and there was no sonographic evidence of involvement of the urinary bladder, uterus, or other adjacent pelvic organs. These findings were consistent with an Altman type II sacrococcygeal teratoma.

Plain radiography was not performed, and advanced imaging modalities such as MRI or CT were unavailable due to financial constraints. Nevertheless, the ultrasound findings provided sufficient information for surgical planning.

Baseline laboratory investigations were within normal limits: white blood cell count was 13.28×10^9/L, haemoglobin level was 12.3 g/dL, and platelet count was 421×10^9/L. With a presumptive diagnosis of a mature sacrococcygeal teratoma, the decision was made to proceed with elective surgical excision under general anaesthesia.

2.1. Surgical Intervention

Following thorough preoperative evaluation by the anaesthesia team, the infant was deemed fit for general anaesthesia in accordance with hospital protocol. She was kept nil per os (NPO) for six hours before surgery. Under general anaesthesia, the patient was positioned prone with a soft pad placed under the pelvis to elevate the sacral region for optimal exposure.

Using aseptic technique, a transverse posterior sacral incision was made just superior to the palpable mass. Careful dissection was carried out to expose the underlying structures. Intraoperatively, the mass was identified as a predominantly intrapelvic lesion with both solid and cystic components. It was located beneath the sacrococcygeal fascia and was found to be firmly adherent to both the posterior rectal wall and the coccyx.

Although the tumour had significant attachment to the rectum, there was no gross evidence of mucosal invasion or intraluminal extension. During dissection, the mass ruptured, resulting in the spillage of cystic contents into the operative field. This complicated the procedure and necessitated meticulous irrigation, suctioning, and cautious redefinition of dissection planes. Sharp dissection was employed to carefully mobilize the tumour from the rectum, preserving the integrity of the rectal wall and avoiding injury to adjacent pelvic structures.

Given the firm attachment to the coccyx and the risk of recurrence if any residual tissue remained, a coccygectomy was performed to ensure complete tumour excision with clear margins. Haemostasis was achieved, the surgical site was irrigated thoroughly, and the wound was closed in layers using Vicryl 2/0 and 3/0 sutures. The infant tolerated the procedure well and was transferred to the recovery unit in stable condition.

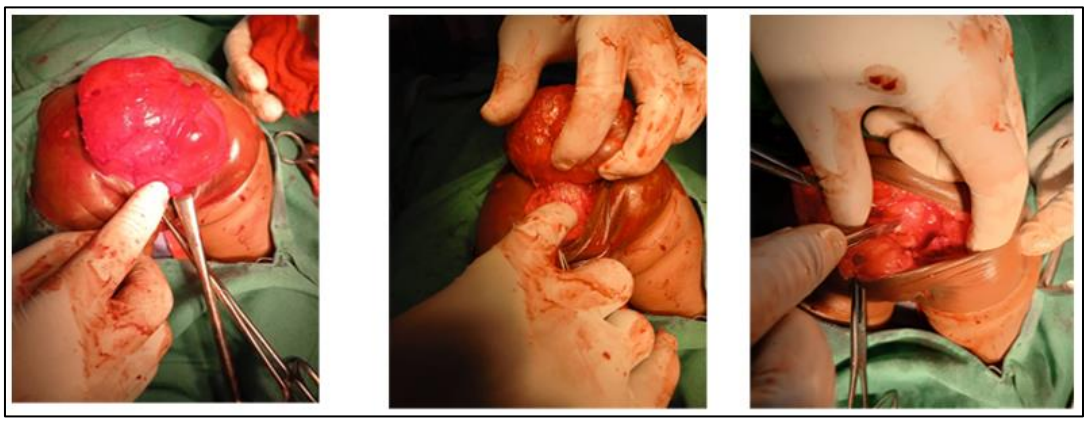

Figure 2 Progression of excision surgery

2.2. Postoperative Course

The postoperative period was uneventful. The surgical incision was clean and healing well, as demonstrated in Figure 5. The infant remained admitted to the surgical ward for three days, during which she received intravenous antibiotics (ceftriaxone–sulbactam 250 mg once daily) and pain management with intravenous paracetamol (75 mg every 6 hours for three days).

Bowel function resumed within 48 hours post-surgery, and the mother noted a marked improvement in defecation frequency compared to the preoperative state. The infant tolerated feeds well and showed no signs of infection or wound complications during the admission period.

Although follow-up had been scheduled for two weeks post-discharge, the patient missed this review due to transportation challenges. She was later seen at six weeks postoperatively. At that time, the surgical wound had healed satisfactorily, and the infant was thriving with no clinical signs of recurrence or complications.

The patient continues on routine outpatient follow-up. However, serum alpha-fetoprotein (AFP) testing for postoperative surveillance could not be performed due to financial limitations.

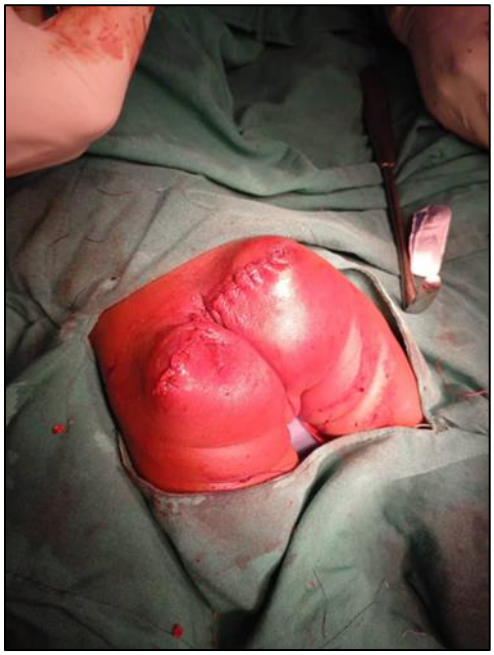

Figure 3 Successful excision and wound closure

2.3. Histopathology

Histological analysis confirmed a mature sacrococcygeal teratoma, composed of well-differentiated tissues from all three germ layers. Identified components included respiratory-type epithelium, hyaline cartilage, glandular structures, adipose tissue, and skeletal muscle. No immature or malignant elements, such as neuroepithelial tissue, were observed. The cauterized surgical margins were free of tumour involvement, indicating complete excision.

3. Discussion

Sacrococcygeal teratomas (SCTs), while commonly benign, can cause significant morbidity due to local mass effects, particularly in delayed or predominantly intrapelvic presentations. In this case, progressive enlargement of the tumor led to anterior displacement of the anus and increased stool frequency—clinical signs that underscore the importance of early evaluation and intervention [5].

Imaging plays a pivotal role in defining the extent of disease and guiding surgical strategy. Although advanced imaging modalities such as MRI or CT were not available in this setting, ultrasound provided valuable information to classify the tumor as Altman Type II and facilitated preoperative planning [6].

Complete surgical excision remains the gold standard in SCT management, with coccygectomy being an essential component. Failure to remove the coccyx significantly increases the risk of recurrence and potential malignant transformation. In cases like this, where the tumor is firmly adherent to the rectum, meticulous dissection is critical to preserve anorectal function and avoid injury to adjacent structures [7,8,9].

4. Conclusion

This case illustrates the importance of early recognition and comprehensive surgical management of SCTs, even in resource-limited settings. In neonates presenting with perineal masses or anorectal distortion, prompt imaging and surgical referral are essential. Coccygectomy should always be performed to ensure complete resection and reduce the risk of recurrence. Successful outcomes can be achieved with careful planning, timely intervention, and adherence to sound surgical principles.

Compliance with ethical standards

Disclosure of conflict of interest

The authors do not have conflict of interest.

Statement of ethical approval

Assent was sought from the parents of the baby.

Statement of informed consent

All participants provided consent to participate in the study.

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