



RESEARCH ARTICLE

SACROCOCCYGEAL TERATOMA IN A LIMITED-RESOURCE SETTING: A CASE REPORT

Felix Pius Omullo (MBChB), Allan Kariuki Ng'ang'a (MBChB), Patrick Osiri (MBChB), Allan Njoroge (MBChB) and Thomas Kimanzi Kitheghe (MBChB)

Department of Obstetrics and Gynecology, Murang'a County Referral Hospital, P.O. Box 69-10200

ARTICLE INFO

Article History

Received 20th September, 2024
Received in revised form
16th October, 2024
Accepted 27th November, 2024
Published online 29th December, 2024

Keywords:

Sacrococcygeal Teratoma, Antenatal Diagnosis, Resource-Limited Settings, Obstetric Complications, Neonatal Outcomes.

*Corresponding author:
Felix Pius Omullo

ABSTRACT

Introduction: Sacrococcygeal teratoma (SCT) occurs commonly in neonates and infants and poses significant obstetric and fetal challenges, especially in resource-strained settings. SCT affects 1 in every 35,000–40,000 live births. Prompt diagnosis and effective management of SCT improve obstetric and neonatal outcomes. **Case presentation:** A 25-year-old gravida 2 para 1+0 at 27 weeks gestation was admitted to the hospital with complaints of intermittent lower abdominal pain and reduced fetal movements. Physical examination revealed bilateral pitting oedema up to the knees and a fundal height of 38 weeks, suggesting a large-for-gestational-age (LGA) fetus. Obstetric ultrasonography showed a single live fetus in cephalic presentation with a sacrococcygeal mass measuring 11.44 × 9.87 × 12.70 cm and mild polyhydramnios. Biochemical investigations were normal. After stabilisation and counselling, the patient was referred to a tertiary facility for advanced obstetric, oncology and surgical management. **Clinical discussion:** SCT arises from totipotent germ cells and is classified by histology and anatomy. Antenatal ultrasound is the primary diagnostic tool, although CT/MRI is superior in defining tumor extent. Complications such as fetal hydrops and obstructive effects are common. Complete surgical excision remains the definitive treatment, with the prognosis depending on tumor size, malignancy, and recurrence risk. Resource limitations impede timely interventions in such settings. **Conclusion:** This case demonstrates the critical role of antenatal diagnosis and prompt referral in managing SCT. Multidisciplinary coordination and early intervention can improve outcomes, even in resource-constrained environments.

Copyright©2024, Felix Pius Omullo et al. This is an open access article distributed under the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.

Citation: Felix Pius Omullo (MBChB), Allan Kariuki Ng'ang'a (MBChB), Patrick Osiri (MBChB), Allan Njoroge (MBChB) and Thomas Kimanzi Kitheghe (MBChB). 2024. "Sacrococcygeal Teratoma in a Limited-resource Setting: A Case Report", *International Journal of Recent Advances in Multidisciplinary Research*, 11, (12), 10517-10519.

INTRODUCTION

SCT is the most common tumor in the neonatal period (1,2). It is located at the base of the coccyx and comprises numerous tissue types. It is postulated that it originates from embryonic germ cell layers (3). Despite being a common neoplasm among neonates and infants, SCT has been reported in adults (2). Most SCTs are cystic and benign, and only 1-2% are malignant (4). Due to the prominent vascularity of some solid SCTs, complications often arise in the perinatal period (3). The morbidity and mortality associated with SCT are attributed to dystocia from tumor masses, preterm delivery, and suboptimal fetal development (5). These complications often worsen obstetric outcomes. SCT presents significant diagnostic and management challenges in resource-limited settings where advanced imaging and surgical expertise may be scarce (4). Early antenatal diagnosis and timely referral to higher-level care are critical in such environments to mitigate complications and optimise maternal and neonatal outcomes.

This case calls attention to a rare case of SCT, prompting an exploration into antenatal diagnosis, gaps in care in resource-limited facilities, and the need for timely referral.

Case Presentation

A 25-year-old gravida 2 para 1+0 at 27 weeks gestation presented with intermittent lower abdominal pain and reduced fetal movements. The pain was sudden in onset, non-radiating, and not worsening in intensity or frequency, occasionally subsiding and reappearing. She denied taking over-the-counter medications, experiencing fever, liquor drainage, or changes in bowel or urinary habits. She had attended 4 regular ANC visits at another peripheral facility. On examination, she had bilateral pitting edema up to the knees, non-tender. Abdominopelvic examination showed a fundal height of 38 weeks, suggesting a large-for-gestational-age (LGA) fetus. An obstetric ultrasound revealed a single live fetus in cephalic presentation with a well-defined isoechoic mass in the sacrococcygeal region measuring 11.44 × 9.87 × 12.70 cm

(Figure 1). The estimated fetal weight was 1538 g, corresponding to 29 weeks gestation. Doppler assessment of the lesion showed low color flow, with no features of vascular compromise or fetal hydrops. Additional findings included mild polyhydramnios, with the deepest liquor pool of 8.3 cm. The biophysical profile (BPP) score was 8/8. Fetal parameters (cardiac activity, femur length, and head circumference) were within normal limits and matched the gestational age. Moreover, all biochemical investigations (CBC, RFTs, UECs, blood glucose levels, urinalysis, PT/INR, and ECG) were normal. After further consultations, the patient was counselled on the possible complications associated with SCT and prognosis and referred to Kenyatta National Hospital (KNH) for advanced management and surgical care.



Figure 1. The ultrasound 2D image at 27 weeks of gestation showing a well-defined isoechoic mass in the sacrococcygeal region

DISCUSSION

Teratomas are the most common germ cell tumors, comprising tissues derived from the three germ cell layers (endoderm, mesoderm, and ectoderm) (2). The sacrococcygeal, mediastinal, and gonadal regions are the common sites for teratomas (2,3). Histopathologically, teratomas are classified as immature and mature (2,6). Mature teratomas make up 75-80%. Specifically, SCTs can be classified depending on their anatomic locations (3). According to the Altman classification system, type I tumors are predominantly external (45%), type II are external with a significant intrapelvic segment (35%), type III are mainly intrapelvic (10%), and type IV are exclusively internal tumors (10%) (3). Type I tumors have the lowest risk of malignancy (2). Despite antenatal sonography being the most used modality in making diagnoses, CT and MRI remain superior in determining SCT's origin, extent, and relationship to the pelvic and abdominal organs (7).

SCTs are believed to arise from the remnant of the primitive streak in the coccyx during the early third week of gestation from the totipotent cells of Hensen's code (3). 62% of SCTs comprise both cystic and solid elements (3). The cystic teratomas are often filled with serous fluids, mucoid, or sebaceous constituents and are lined with true epithelium (8). SCTs usually present during the gestation period of between the 22nd and 34th week of gestation (9). When diagnosed by an antenatal ultrasound, SCT is linked with perinatal complications (10). Clinical manifestations of SCT depend on the extent and location of the neoplasm (11). Most patients present with low back pain, urinary and bowel symptoms, and

lower limb swelling, which are all attributed to the neoplasm's mass effect (3, 11). The patient presented with bilateral, non-tender lower limb pitting edema in this case. Tumor markers such as AFP, CEA, and HCG are markedly elevated in malignant neoplasms (12). Nevertheless, markers of biochemical origin are not observed in benign types (3). Biochemical investigations were normal in this particular case presentation. Fetal hydrops is one of the significant complications of SCT (13). When hydrops occur, the mortality rate increases two-fold (3,14). Hydrops is caused by high-output cardiac failure and anemia secondary to intramural hemorrhage (14,15). In scenarios where fetal hydrops are observed, prenatal monitoring is invaluable. Significantly large SCT exerts pressure on adjacent structures, culminating in obstructive hydronephrosis, hip dislocation, and anorectal displacement (3).

The other anomalies witnessed with SCT are cleft lip and palate, hydrocephalus, spina bifida, and transposition of great vessels (3,16). Complete surgical resection is the standard treatment for SCT (17,18). More importantly, it should be performed promptly to avoid tumor rupture, hemorrhage, recurrence, or malignant transformation (18). In this case, the patient was referred to KNH at 29 weeks of gestation after being stabilised. Altman type I and II are operated via the posterior approach, whereas large type III often requires transabdominal (anterior) and posterior approaches (19). Laparoscopic management is considered for type IV SCT (18). Coccygectomy prevents tumor tissue spillage during surgery (19). The prognosis of SCT is favorable (3,4,17,18,19). However, the condition may recur (10-15%), so careful monitoring with DRE and serum AFP after every 3 months is recommended (18). Poor prognostic factors are older age at diagnosis (>2 years), solid components within the neoplasm, and malignant histology (18,20). Fetal treatment of SCT, open or interventional, has not demonstrated better outcomes. However, these approaches are often considered in pregnancies with complications such as fetal hydrops, high-output cardiac failure, and preterm deliveries.

CONCLUSION

Antenatal diagnosis of SCT in resource-limited settings is critical for timely management and referral. Multidisciplinary collaboration and maternal counseling are pivotal in addressing associated complications and improving prognosis.

ACKNOWLEDGMENT

The authors thank Dr. Tungani Muchiri and Murang'a County Referral Hospital for supporting the preparation of this case report.

Ethical Considerations: The patient gave written informed consent to publish medical details and images.

Author Contributions

Felix Pius Omullo: Lead author, compilation, and final review; Allan Kariuki Ng'ang'a: Review and approvals; Patrick Osiri: Manuscript drafting; Allan Njoroge: Manuscript drafting; Thomas Kimanzi Kitheghe: Review and compilation

Source of Funding: No funding was received for this case report.

Competing Interests: The authors declare no competing interests.

REFERENCES

- Swamy R, Embleton N, Hale J. Sacrococcygeal teratoma over two decades: Birth prevalence, prenatal diagnosis and clinical outcomes. *Prenatal Diagnosis*. 2008 Nov;28(11):1048–51.
- Cui S, Han J, Khandakar B, Friedman B, Nunez D, Sara GA, et al. Recurrent Adult Sacrococcygeal Teratoma Developing Adenocarcinoma: A Case Report and Review of Literatures. Nagashima Y, editor. *Case Reports in Pathology* [Internet]. 2021 Nov 27 [cited 2023 Feb 27];2021:1–6. Available from: <https://www.ncbi.nlm.nih.gov/pmc/articles/PMC8643248/>
- Shrestha HK, Shrestha RG. Sacrococcygeal Teratoma: A Case Report. *Journal of Nepal Medical Association*. 2020 Jul 31;58(227).
- Keslar PJ, Buck JL, Suarez ES. Germ cell tumors of the sacrococcygeal region: radiologic-pathologic correlation. *RadioGraphics*. 1994 May;14(3):607–20.
- Vogl D, Riel K. Präsakraler Tumor mit sakraler Meningozele. *European Journal of Pediatric Surgery*. 1988 Oct;43(05):361–4.
- Romagnani E. Mediastinal mature teratoma with an immature component--what about the treatment? *Annals of Oncology*. 2006 Mar 17;17(10):1602–4.
- Guo JX, Zhao JG, Bao YN. Adult sacrococcygeal teratoma: A review. *Medicine*. 2022 Dec 30;101(52):e32410.
- Julián Eduardo Grisales-Gallo, Salinas-Castro KJ, Nasner D, Toro-Gutiérrez JS. Mature cystic teratoma overlapping with giant serous cystadenoma of the ovary: A case report. *Radiology Case Reports* [Internet]. 2023 Nov 2 [cited 2024 Dec 1];19(1):218–22. Available from: <https://pmc.ncbi.nlm.nih.gov/articles/PMC10651425/>
- Hassan HS, Elbatarny AM. Sacrococcygeal teratoma. *Annals of Pediatric Surgery*. 2014 Jul;10(3):72–7.
- Savukyne E, Krzconaviciute S, Vaitkeviciute M, Machtejeviene E, Rubaviciute I. Antenatal and histological diagnostics of cystic sacrococcygeal teratoma. *Clinical case and literature review. Case Reports in Perinatal Medicine*. 2023 Jan 1;12(1).
- Zheng XQ, Yan JY, Xu RL, Wang XC, Chen X, Huang KH. A Clinical Analysis of the Diagnosis and Treatment of Fetal Sacrococcygeal Teratomas. *Cancer Management and Research*. 2020 Dec;Volume 12:13185–93.
- Rath A, Mitra S, Adhya AK, Majumdar SKD. Immature teratoma with very high AFP levels. *European Journal of Obstetrics & Gynecology and Reproductive Biology: X*. 2022 Dec;16:100170.
- Abiad M, Nikan Zargarzadeh, Javinani A, Krispin E, Shamshirsaz AA. Fetal Teratomas: Advances in Diagnosis and Management. *Journal of Clinical Medicine*. 2024 Oct 19;13(20):6245–5.
- Van Mieghem T, Al-Ibrahim A, Deprest J, Lewi L, Langer JC, Baud D, et al. Minimally invasive therapy for fetal sacrococcygeal teratoma: case series and systematic review of the literature. *Ultrasound in Obstetrics & Gynecology*. 2014 May 8;43(6):611–9.
- Nagy A, Roger. *Fetal Hydrops*. Springer eBooks. 2022 Jan 1;305–34.
- Yoon HM, Byeon S, Hwang JY, Kim JR, Jung AY, Lee JS, et al. Sacrococcygeal teratomas in newborns: a comprehensive review for the radiologists. *Acta Radiologica*. 2017 May 22;59(2):236–46.
- Xu X, Zhao F, Cheng X, Zhong W, Liu J, Jiang W, et al. Adult sacrococcygeal teratoma: a retrospective study over eight years at a single institution. *Journal of Zhejiang University Science B*. 2019 Jul 10;20(8):670–8.
- Phi JH. Sacrococcygeal Teratoma: A Tumor at the Center of Embryogenesis. *Journal of Korean Neurosurgical Society*. 2021 May 1;64(3):406–13.
- Sinha S, Kumar Sarin Y, P Deshpande V. Neonatal sacrococcygeal teratoma: our experience with 10 cases. *Journal of neonatal surgery* [Internet]. 2013 Jan;2(1):4. Available from: <https://pubmed.ncbi.nlm.nih.gov/26023424/>
- Farzana Pashankar, Murray MJ, Gell J, MacDonald N, Shamash J, Billmire DF, et al. Consensus and controversy in the management of pediatric and adult patients with ovarian immature teratoma: the Malignant Germ Cell International Consortium perspective. *EClinicalMedicine*. 2024 Mar 1; 69:102453–3.
