ORIGINAL RESEARCH PAPER
Volume - 14 | Issue - 05 | May - 2025 | PRINT ISSN No. 2277 - 8179 | DOI : 10.36106/ijsr
INTERNATIONAL JOURNAL OF SCIENTIFIC RESEARCH

SACROCOCCYGEAL TERATOMA WITH BOWEL DUPLICATION: A CASE REPORT

Paediatric Surgery

Dr Pratiksha Basant Joshi	Senior Resident, Department of Pediatric Surgery, Lokmanya Tilak Municipal Medical College and Hospital, Sion, Mumbai, India.
Dr Nandini Desai	Senior resident, Department of Pediatric Surgery, Lokmanya Tilak Municipal Medical College and Hospital, Sion, Mumbai, India.
Dr Paras Kothari	Professor & Head, Department of Pediatric Surgery, Lokmanya Tilak Municipal Medical College and Hospital, Sion, Mumbai, India.
Dr Abhaya Gupta	Additional Professor, Department of Pediatric Surgery, Lokmanya Tilak Municipal Medical College and Hospital, Sion, Mumbai, India.
Dr Shahaji Deshmukh	Assistant Professor, Department of Pediatric Surgery, Lokmanya Tilak Municipal Medical College and Hospital, Sion, Mumbai, India.

ABSTRACT

Sacrococcygeal teratoma (sct) is the most common neoplasm in fetus and newborn. Sometimes, mature sacrococcygeal teratomas can be highly differentiated and SCT with organoid differentiation are rarely reported in the literature1. Here we take the opportunity to report a case of mature sacrococcygeal teratoma with the uncommon finding of presence of duplex small bowel within the mass. Neonate with type 2 sacrococcygeal teratoma was referred to our hospital. The mass was excised with gross specimen suggestive of bowel duplication which was confirmed on histopathological examination. Child is thriving well on follow-up.

KEYWORDS

Sacrococcygeal Teratoma, Duplex Small Bowel

INTRODUCTION

Sacrococcygeal teratoma are the most common extragonadal tumor in neonates accounting upto 70% of all teratomas in children. They are more common in girls (4:1) but are found more malignant in boys. Newborn present with sacral mass ,many detected in prenatal ultrasonography. Older children present with symptoms related to compression of bladder or rectum. Rate of malignancy increases with age. AFP levels should be obtained and followed to ensure that they return to normal by 9 months of age. Imaging can help to guide the degree of pelvic and abdominal involvement.

Case Presentation

Day of life 2 female child weighing 3.5 kg delivered at 42 weeks by LSCS referred with mass in sacral region since birth. ANC anomaly scan at 7th month showed sacrococcygeal teratoma. Mass of size approximately 10 x 8 cm was present in sacral region which was skin covered with visible dilated veins. The neonate was otherwise asymptomatic and had no associated spinal anomaly.

Routine preoperative investigations were within normal range. Beta HCG <0.2 mIU/ml, AFP – 11412ng/ml. MRI was s/o cystic sacrococcygeal teratoma type II with intra and extrapelvic component. Ileal loops with mesenteric fat seen in the extrapelvic component. Mass was excised completely along with coccyx and excess skin was removed.

Fig.2: Duplication Of Small Bowel[1] Seen

Gross specimen findings were solid cystic mass of approximately 6x4 cm with irregular surface with areas of congestion with two tubular structures approximately 1 and 3 cm each noted.

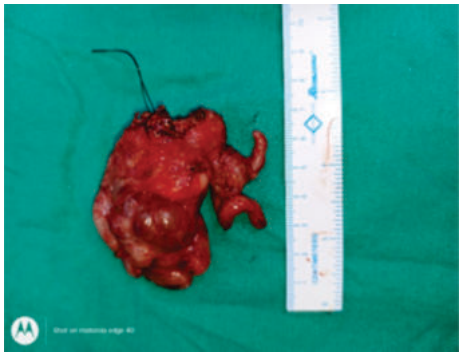

Fig.3: Gross Specimen Of Sacrococcygeal Teratoma With Duplication Of Small Bowel

Postoperatively patient was managed with nutritional support and discharged after stabilisation. On follow-up patient had no complaints.

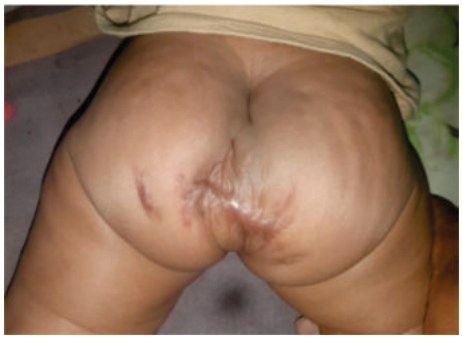

Fig.4: Postoperative Scar On Followup

Histopathology was suggestive of benign germ cell tumor - mature cystic teratoma with all three mature components – ectodermal, endodermal, mesodermal origin with small bowel mucosa. No e/o immature glial tissue.

DISCUSSION

Sacrococcygeal teratomas (SCT) are composed of diverse tissues foreign to the anatomical site of origin. Incidence being 1–2 per 40,000

deliveries. Totipotent cells from Hansen's node or primitive germ cells are considered to be the origin of SCT[2].

SCT are derived from all three germinal layers and contain neural elements, squamous and intestinal epithelium, skin appendages, teeth and, at times, calcium. A spectrum of anomalies can be seen in neonates ranging from conjoined twins to external parasitic twins to fetus in fetu to "fetiform"[3] teratomas to well-differentiated teratomas depending on the timing of embryological insult and degree of differentiation. Their inheritance may be sporadic but, occasionally, autosomal dominant. They can also be associated with other congenital anomalies like urogenital[4], Currarino's triad[5].

The American Association of Pediatric Surgery has classified them into four types[6]:
(1) 47%, exophytic and external;
(2) 34%, dumbbell shaped, with equal internal/external components (present case);
(3) 9%, primarily located within the abdomen/pelvis;
(4) 10%, entirely internal, no external components.

Intrauterine surgery by interrupting the feeding vessel has been suggested when the fetus develops hydrops prior to viability. The role of open intrauterine surgery, however, is limited because it carries high morbidity to the mother and the fetus. Radiofrequency ablation has shown promising results in the treatment of fetal SCT[7].

In a viable foetus, caesarean section delivery is recommended if SCT measures >5 cm to prevent dystocia or rupture of SCT. Early excision of the tumor in continuity with the coccyx is recommended as failure to remove the coccyx results in 30–40% risk of local recurrence[8]. Excess or damaged skin is removed to allow a cosmetic reconstruction. Combined abdominoperineal approach may be required in type 2/3 SCT.

Risk of malignancy depends on the time of diagnosis (7–10% at <2 months, 37% at 1 year and 50% at 2 years). In benign tumors, disease-free survival is >90%, whereas malignant tumors have significant mortality. Malignant tumors[9] require chemotherapy and radiation preoperatively or postoperatively. Hence, frequent follow-up with serum α-fetoprotein level measurement and surveillance by imaging is recommended.

CONCLUSION

This case adds to the literature a rare presentation of sacrococcygeal teratoma exhibiting organoid differentiation(duplication of small bowel). Various tissues have been reported in sct – nephroblastic elements[10], adrenal gland[11], pulmonary tissue[12]. Small bowel is reported in one case[1].This is the first case to report a duplicated bowel as a component of SCT. More number of cases of organoid differentiation needs to be reported in order to establish definite pathogenesis behind such differentiation.

REFERENCES

1. Aslan A, Karagüzel G, Gelen T, Melikoğlu M. Sacrococcygeal teratoma showing organoid differentiation: report of a case. Surg Today. 2003;33(7):560-3. doi: 10.1007/s10595-002-2528-6. PMID: 14507007.
2. Tapper D, Lack EE. Teratomas in infancy and childhood. A 54-year experience at the Children's Hospital Medical Center. Ann Surg. 1983;198(3):398–410. doi: 10.1097/00000658-198309000-00016. [DOI] [PMC free article] [PubMed] [Google Scholar]
3. Sood N, Kamboj M, Chaabra M. Sacrococcygeal fetiform teratoma altman type 1: a rare case report in a 11 year old girl. Indian J Surg. 2013 Jun;75(Suppl 1):359-61. doi: 10.1007/s12262-012-0703-5. Epub 2012 Jul 28. PMID: 24426616; PMCID: PMC3693311.
4. Shalaby MS, O'Toole S, Driver C, Bradnock T, Lam J, Carachi R. Urogenital anomalies in girls with sacrococcygeal teratoma: a commonly missed association. J Pediatr Surg. 2012 Feb;47(2):371-4. doi: 10.1016/j.jpedsurg.2011.11.038. PMID: 22325393.
5. Hu Q, Yan Y, Liao H, Liu H, Yu H, Zhao F. Sacrococcygeal teratoma in one twin: a case report and literature review. BMC Pregnancy Childbirth. 2020 Dec 2;20(1):751. doi: 10.1186/s12884-020-03454-1. PMID: 33267783; PMCID: PMC7709297.
6. Yoon HM, Byeon S, Hwang J-Y, et al. Sacrococcygeal teratomas in newborns: a comprehensive review for the radiologists. Acta Radiologica. 2017;59(2):236-246. doi:10.1177/0284185117710680
7. Litwińska M, Litwińska E, Janiak K, Piaseczna-Piotrowska A, Szaflik K. Percutaneous Intratumor Laser Ablation for Fetal Sacrococcygeal Teratoma. Fetal Diagn Ther. 2020;47(2):138-144. doi: 10.1159/000500775. Epub 2019 Jul 10. PMID: 31291630.
8. Ho KO, Soundappan SV, Walker K, Badawi N. Sacrococcygeal teratoma: the 13-year experience of a tertiary paediatric centre. J Paediatr Child Health. 2011 May;47(5):287-91. doi: 10.1111/j.1440-1754.2010.01957.x. PMID: 21599781.
9. van Heurn LJ, Derikx JPM, Hall N, Aldrink JH, Bailez MM, Chirdan LB, Fumino S, Hesse A, Soyer T, StPeter S, Twisk J, Yang T, van Heurn ELW; SCT-study consortium. Malignant transformation and tumour recurrence in sacrococcygeal teratoma: a global, retrospective cohort study. Int J Surg. 2024 Nov 1;110(11):7177-7186. doi: 10.1097/JS9.0000000000002045. PMID: 39248311; PMCID: PMC11573091.
10. Ma Y, Zheng J, Zhu H, Shen C, Zheng S, Xiao X, Chen L. Sacrococcygeal teratoma with nephroblastic elements: a case report and review of literature. Int J Clin Exp Pathol. 2014 Oct 15;7(11):8211-6. PMID: 25550874; PMCID: PMC4270538.
11. Mandal B, Chatterjee G, Bhattacharya K, Roy D, Das RN, Chatterjee U. Sacrococcygeal teratoma with complete adrenal gland. J Cancer Res Ther. 2015 Oct-Dec;11(4):1040. doi: 10.4103/0973-1482.148690. PMID: 26881658.
12. Gupta B, Yadav S, Khurana N, Raj P. Sacrococcygeal Teratoma in a Seven-Day-Old Child with Pulmonary Differentiation. J Clin Diagn Res. 2017 Aug;11(8):SD01-SD02. doi: 10.7860/JCDR/2017/27273.10357. Epub 2017 Aug 1. PMID: 28969230; PMCID: PMC5620871.