Case Report

Fetiform sacrococcygeal teratoma simulating parasitic twin: A case report

Amna Qaiser*, Hassan Mahmud, Mahboob Ahmad Bhutta, Faryal Ilyas

Department of Pediatric Surgery, University of Child Health Sciences Lahore, Pakistan

Correspondence*: Amna Qaiser, Department of Pediatric Surgery, University of Child Health Sciences Lahore, Pakistan, E-mail: amnaqaiser7194@gmail.com.
 | Submitted on: 18-11-2023 | Accepted on: 01-03-2024
Conflict of interest: None | **Source of Support:** Nil

Cite as: Qaiser A, Mahmud H, Bhutta MA, Ilyas F. Fetiform sacrococcygeal teratoma simulating parasitic twin: A case report. J Child Health Sci. 2024: Ahead

ABSTRACT

Introduction: Sacrococcygeal teratoma and parasitic twinning are distinct anomalies, each requiring different treatments and resulting in varied outcomes. Fetiform teratoma can mimic parasitic twins, posing diagnostic and management challenges.

Case Report: A one-hour-old neonate presented with active bleeding from a large, ruptured mass in the sacrococcygeal region. The mass contained identifiable organs, such as the scalp and the ear. Hemostasis was immediately secured with packing and dressing. MRI and tumor markers were performed to differentiate it from parasitic twins. The fetiform teratoma was excised along with a coccygectomy. Histopathology confirmed it as a mature sacrococcygeal teratoma. Postoperative recovery was uneventful, except for a wound infection that required wound care.

Conclusion: Fetiform teratoma may mimic parasitic twins and requires a thorough investigation for an accurate diagnosis before proceeding with surgery.

Keywords: Sacrococcygeal teratoma, Fetiform teratoma, Parasitic twins, Neonate, Case report

INTRODUCTION

Teratoma is a type of germ cell tumor that mostly occurs in the sacrococcygeal region and is more common in females [1, 2]. On the other hand, parasitic twinning is not a tumor but rather an abnormality of conjoined twins that fails to develop into a separate entity [3]. It is important to note that sacrococcygeal teratoma and parasitic twinning are two distinct anomalies that require different management approaches.

Fetiform teratomas exhibit certain structures that resemble a parasitic twin [4]. They are difficult to distinguish on clinical examination and often require radiological and histopathological examinations for differentiation [4].

Herein, we report a case of a ruptured fetiform sacrococcygeal teratoma. This case is being reported due to the diagnostic and management challenges it posed to the surgical team.

CASE REPORT

An hour-old male baby, delivered via elective cesarean section, presented with bleeding from a large, peculiar-looking mass at the lower back (Fig. 1). The cesarean section was performed in a private facility in the periphery, but the bleeding occurred due to mishandling. An antenatal scan during the 7th month of gestation revealed a heterogeneous mass in the sacral region, but no hydrops were observed. The mother had no history

of diabetes, hypertension, or exposure to teratogens.

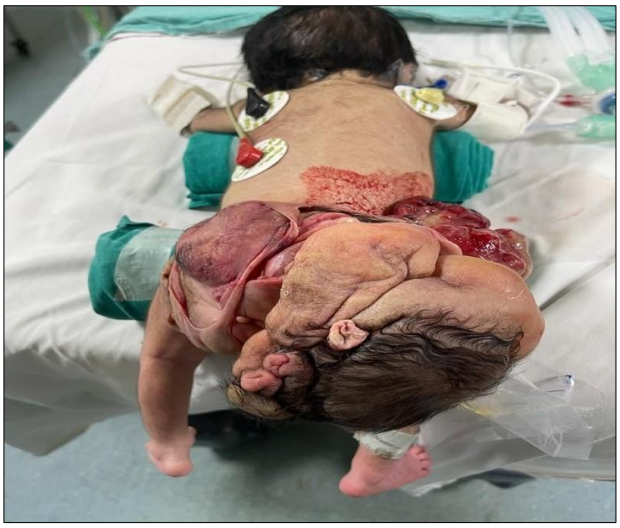

Figure 1: Tumor at sacrococcygeal region showing scalp, hairs, ear, and soft tissue structure.

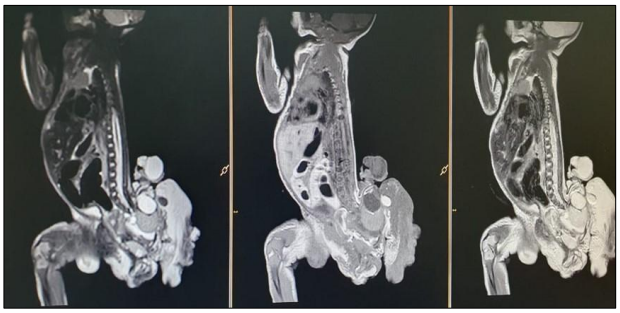

Figure 2: MRI of sacrococcygeal tumor with finding of heterogeneously enhancing pelvic mass lying in pre sacral space, extending in to left hemi pelvis, having endophytic and huge exophytic component with right wall displacement of rectum and extending up to s2 level. No intra spinal extension seen.

Upon examination, the baby appeared pale but otherwise normal. Perineal examination revealed normal male external genitalia and a regular anal opening. A massive complex mass measuring approximately 20x18cm was evident on the sacrococcygeal region, displaying obvious rupture and active bleeding. The mass resembled a parasitic twin, with identifiable structures such as a scalp with hair, ear, and brain tissue. No other spinal or limb deformities were noted.

The baby was promptly transferred to the emergency operating theater, where hemostasis was achieved by packing and dressing the ruptured site of the tumor. Intravenous fluids and blood products were administered due to anemia (Hb 9g/dl) resulting from bleeding. Intravenous tranexamic acid and vitamin K were also given. Since hemostasis was successful, surgical excision was deferred pending further investigation of the nature of the mass.

An MRI of the tumor revealed a large 13x12.2x9.6cm heterogeneously enhancing mass located in the presacral space, extending into the left hemipelvis, with a significant exophytic component indenting the rectal wall but not involving it. This finding was suggestive of Sacrococcygeal teratoma Altman type II (Fig. 2). Alpha-fetoprotein levels were within the normal range for the age. Laboratory tests were conducted privately as they were not available in the hospital at that time, potentially leading to some discrepancies.

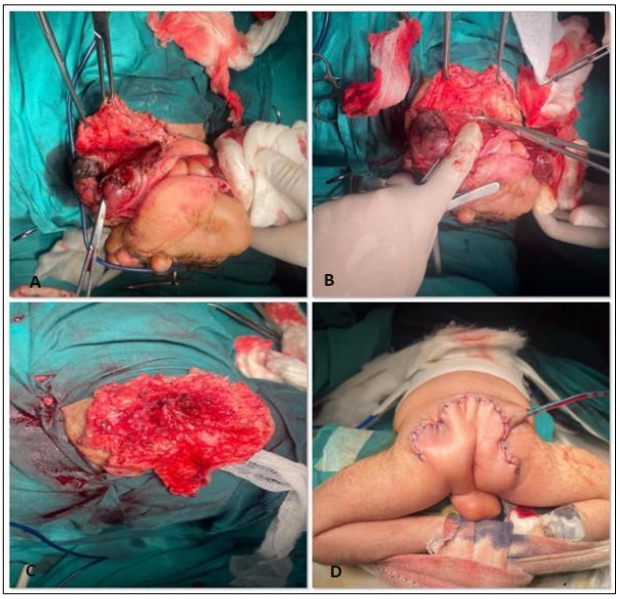

Figure 3: Intraoperative pictures: (a) showing mobilization of normal skin flap from tumor margins, (b) showing ligation of median sacral artery, (c) showing pelvic floor after complete excision of tumor, (d) showing construction of pelvic floor muscles.

After optimization and obtaining informed consent, the baby underwent excision of the mass on the elective surgical list. A large chevron incision was made in the jack-knife position. Identification of the tumor planes was challenging as the margins of normal skin and the tumor were fused in some areas. Complete excision of the tumor was achieved, along with coccygectomy. Operative findings revealed a ruptured, exophytic mass measuring about 17x17cm, containing scalp hair, cartilage, and a malformed ear. It was adherent to the coccyx with pelvic extension, displacing the anal canal and rectum but not involving them (Fig. 3). Pelvic floor reconstruction was performed, and the wound was closed. Nerve stimulations indicated normal anal sphincter contractions.

Postoperative recovery was uneventful except for a wound infection that developed on the 3rd postoperative day, which was managed with antiseptic dressings and wound care. The patient was discharged on the 10th postoperative day in good clinical condition.

Histopathology of the tumor revealed it as a mature cystic teratoma, displaying stratified squamous epithelium with underlying skeletal muscles, fat, cartilage, mucinous glands, and brain tissue (Fig. 4).

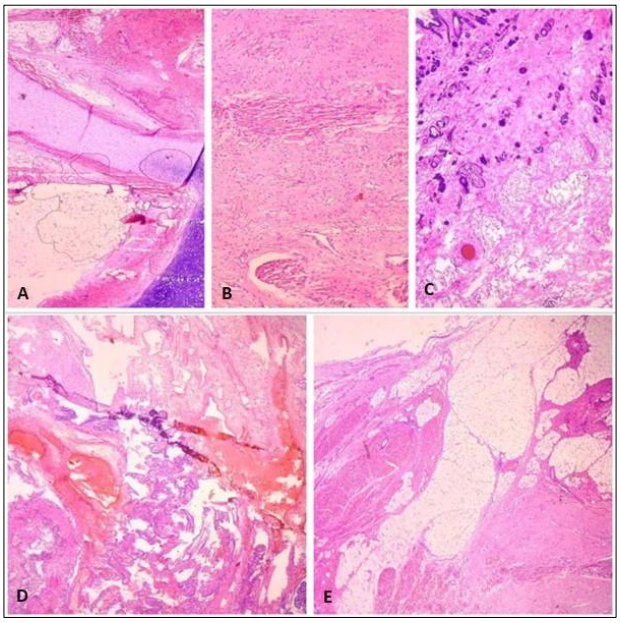

Figure 4: Histopathology: (a) showing fat and cartilage, (b) showing glial tissue and skeletal muscle, (c) showing dermal appendages, (d) showing few papillary structures, (e) showing skeletal muscle.

The baby was followed up at 3 months and showed no signs or symptoms of recurrence. There were no indications of bladder or bowel dysfunction.

DISCUSSION

Sacrococcygeal teratomas, although rare, rank among the most common tumors affecting newborns. While predominantly benign and mature, cases of immature and malignant teratomas have also been well-documented in the literature [2]. These benign tumors, typically located in the sacrococcygeal region, are believed to originate from the abundance of pluripotent cells in the caudal region, with timely diagnosis and intervention generally leading to favorable outcomes [2].

Derived from germinal layers, sacrococcygeal teratomas encompass a diverse array of tissues originating from ectoderm, mesoderm, and endoderm, including elements such as hair, bones, cartilage, muscle, and intestine [2]. In certain instances, these tissues organize to such an extent that they closely mimic the structure of an organ [4, 5]. These teratomas, termed fetiform teratomas, often exhibit a striking resemblance to parasitic twins, showcasing the intriguing spectrum of development within these unique teratological entities [4, 5].

Distinguishing fetiform teratomas from parasitic twins is crucial, as the former can be malignant, whereas the latter is not considered a tumor. The management approach for sacrococcygeal teratomas (SCT) and parasitic twinning at this site significantly differs. For SCT, the standard management protocol includes diagnostic imaging studies and tumor marker assessments [6]. Subsequently, the recommended course of action typically involves surgical excision, including coccygectomy, to ensure complete tumor removal [2, 6]. In contrast, for parasitic twinning in the sacrococcygeal region, tumor markers are not necessary, as parasitic twinning is not considered a tumor. However, due to the potential involvement of the spine and spinal cord in parasitic twins, a collaborative effort often involves a pediatric neurosurgeon [7].

Large sacrococcygeal teratomas can present challenges at birth, particularly when complicated by iatrogenic injuries [1], as observed in the index case. The standard approach for bleeding and ruptured sacrococcygeal teratomas involves emergent management, which includes controlling bleeding and excising the tumor during the same procedure [8]. However, in this specific case, due to diagnostic uncertainty, immediate excision was deferred. Instead, the focus was on controlling bleeding, allowing time for comprehensive imaging and tumor marker workup. This cautious strategy proved effective, enabling a preoperative diagnosis of the condition as a fetiform Altman type II sacrococcygeal teratoma. Subsequently, a successful surgical intervention was carried out, involving complete tumor excision along with coccygectomy.

CONCLUSION

Fetiform teratomas may simulate parasitic twins and pose diagnostic and management challenges. This case underscores the importance of a tailored and comprehensive diagnostic approach in managing complex sacrococcygeal ter-

atomas, ensuring optimal decision-making and successful outcomes.

Consent to Publication: Author(s) declared taking informed written consent for the publication of clinical photographs/material (if any used), from the legal guardian of the patient with an understanding that every effort will be made to conceal the identity of the patient, however it cannot be guaranteed.

Authors Contribution: Author(s) declared to fulfill authorship criteria as devised by ICMJE and approved the final version. The authorship declaration form, submitted by the author(s), is available with the editorial office.

Acknowledgments: None

REFERENCES

1. Ajaj OA. Ruptured giant sacrococcygeal teratoma. J Neonat Surg. 2022;11:17.
2. Gharpure V. Sacrococcygeal teratoma. J Neonat Surg. 2013; 2:28.
3. Rattan KN, Singh J, Dalal P, Sonika P, Rattan A. Sacral rachipagus parasite: A case report. J Neonat Surg. 2016; 5:16.
4. Kattepura S, Sahadev R, Munianjanappa N, Shankar G, Santhanakrishnan R. Fetiform sacrococcygeal teratoma. Arch Dis Childhood-Fetal Neonat Ed. 2012; 97:F278.
5. Sood N, Kamboj M, Chaabra M. Sacrococcygeal fetiform teratoma altman type 1: A rare case report in a 11 year old girl. Ind J Surg. 2013; 75:359-61.
6. Phi JH. Sacrococcygeal teratoma: a tumor at the center of embryogenesis. J Korean Neurosurg Soc. 2021; 64:406-13.
7. Nadeem MM, Ijaz L, Rehman LU, Hassan MU. Vestigial accessory limbs with spina bifida: Our 5–year experience. Pak J Neurol Surg. 2016; 20:187-91.
8. Alani MA. Successful postnatal management of ruptured giant sacrococcygeal teratoma. J Neonat Surg. 2017; 6:37.