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Sacrococcygeal Teratoma a Rare Tumor in Children: Case Report

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Abstract

Sacrococcygeal teratomas (SCTs) are uncommon congenital tumors that typically develop in newborns, they are rarely associated with chromosomal abnormalities or other congenital anomalies. The majority of pediatric teratomas are benign in the neonatal age group, but the risk of malignancy increases with age. Diagnosis is based on a combination of clinical, radiological, and hormonal findings, but confirmed by anatomopathological study. Treatment is primarily surgical, with the aim of achieving complete resection to prevent recurrence. We present the case of a 22-month-old child who was admitted for management of a sacrococcygeal mass and was diagnosed with an immature teratoma.

Keywords

Sacrococcygeal Mass, Alpha-Fetoprotein, Surgery

1. Introduction

Sacrococcygeal teratoma (SCT) is the rarest tumor, occurring in 1 in 35,000 to 40,000 births [1] [2]. It can be diagnosed antenatally at birth, or later. It is more common in females than in males, accounting for 3 to 4 times. It is uncommon in the pediatric age group, where it accounts for approximately 3% of cancers in children under the age of 15 [3]. Sacrococcygeal teratomas occur from totipotent cells of Hensen's ganglion and contain tissue derived from more than one germ layer, and they are classified into four anatomical types based on the intra- and extra-pelvic extension of the tumor mass [4]. Associated congenital anomalies are observed in 15% - 30% of patients with SCT [5]. The most common presentation of sacrococcygeal teratomas in children is a sacrococcygeal mass [5].

We report the case of an immature hypersecretory teratoma revealed by a sa-

crococcygeal mass at the age of 22 months, and also review the literature to discuss the importance of prenatal diagnosis, early and appropriate management to avoid recurrence, and the importance of subsequent follow-up to monitor for sequelae.

2. Clinical Observation

We present the case of a 22-month-old child female, from a consanguineous marriage, with no notable pathological antecedents; there was no similar case in the family. She was only the daughter of her family resulting from a poorly monitored pregnancy with notion of a single antenatal ultrasound returned without particularity. The history revealed a small sacrocygeal mass detected at birth but ignored by the mother and the medical professional. At the age of 18 months, the mother noticed a rapidly progressive increase in the volume of the mass.

Clinical examination revealed a sacral tumefaction measuring 12 cm × 11 cm, painful on palpation with no ulceration or fistula opposite (**Figure 1**). There was no functional impotence of either lower limb, no any other accompanying signs. There was no fever or change in general condition, and the rest of the somatic examination was unremarkable.

Abdominal and pelvic CT scan showed mass invading the right perineal and gluteal region, suggesting a sacro coccygeal teratoma (**Figure 2**).

Biologically: a hormonal work-up was carried out, showing a very high level of alpha fetoprotein (AFP) at 113,700 ng/ml, while the chorionic gonadotropin hormone (HCG) was negative. The rest of the biological work-up was without anomalies.

An extension work-up revealed a secondary pulmonary mass with multiple bilateral pulmonary nodules and suspicious right internal iliac adenopathy. A pre-chemotherapy work-up was carried out with no abnormalities, then neoadjuvant chemotherapy was started with good clinical progression by reduction in the size of the mass (**Figure 3**), and the level of AFP was decreasing with control after each course of chemotherapy until negativation at 3 ng/ml after the 6th course (**Figure 4**). Radiologically, the reduction in the size of the tumour mass was estimated at 66% compared with the initial size.

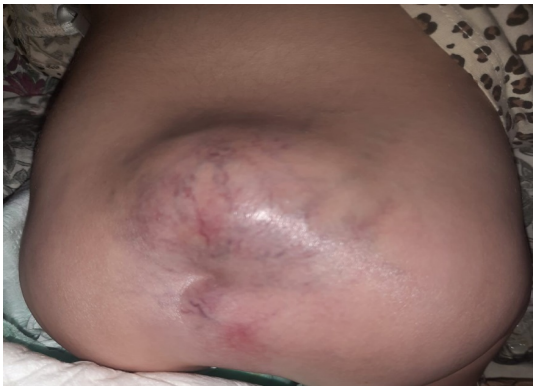

Figure 1. Median sacro coccygeal mass.

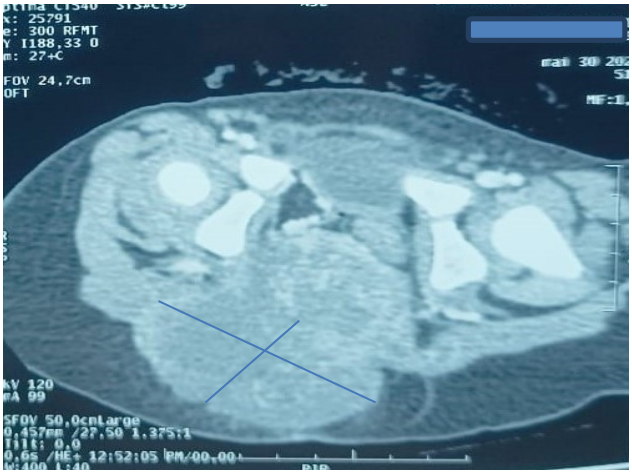

Figure 2. Scanned image showing a sacro coccygeal mass suggestive of a teratoma.

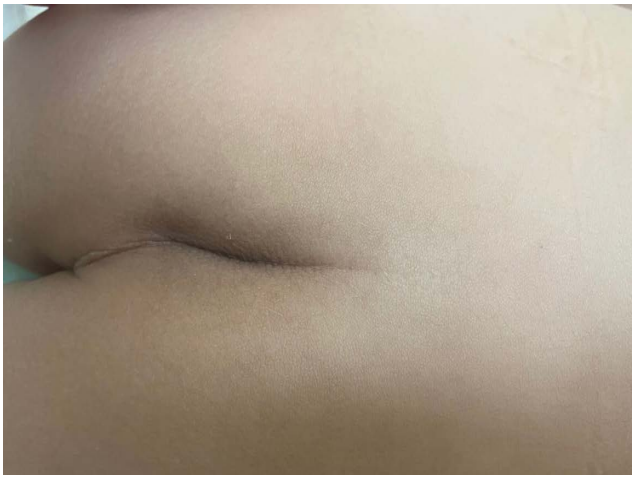

Figure 3. Clinical evolution after 6 courses of neoadjuvant chemotherapy.

Figure 4. Kinetics of alpha fetoprotein (AFP).

A pediatric surgery consult was also conducted, and the child was transferred to the pediatric surgery ward for the complete resection of the tumor and coccyx.

3. Discussion

Teratomas are tumors that arise from pluripotent cells and consist of various tissues that represent all three layers of germ cells [6]. The sacrococcygeal area is the most common extragonadal site, occurring at an incidence of 1/40,000 births, with a female predominance of 3:1 to 4:1 in female-to-male [6].

The early growth of this tumor is largely unknown from an embryological perspective, but it most likely originates from an unorganized growth of concentrated mesodermal cells that appear as early as 18 days and are located in the sacrum and coccyx area, where they are joined by parts of the migrating Henson's node [7]. Nevertheless, in neonates and young infants, the sacrococcygeal area is the most common site for germ cell tumors (GCT) development; SCT develops from the sacrum and coccyx, protruding outwards and growing into the pelvic cavity [8].

After a thorough examination, 15% - 30% of patients with SCT have associated congenital anomalies. The most common anomalies in the urogenital system are hydronephrosis, which can be thought of as a result of compression by the tumor mass. This percentage of congenital anomalies is higher than that of the normal population (3% - 4%), but there are very few causative associations, with the exception of the extremely rare Currarino triad, which is made up of a presacral mass, anorectal anomalies, and sacral bony defects [8].

The Altman [5] classification system categorizes SCT into four types based on their anatomical location: type I, which are primarily external tumors (45%); type II, which present externally but with a significant intrapelvic portion (35%); type III, which are primarily intrapelvic tumors (10%); and type IV, which are presacral tumors without an external component (10%). SCT that are classified as large type II-IV can have mass effects on intrapelvic organs and present with severe issues like constipation, fecal incontinence, and urinary incontinence. Large type III SCT frequently requires extensive abdomino-sacral resection and carries a high risk of a poor functional outcome [8].

In addition, cross-sectional imaging studies like CT scan and magnetic resonance imaging (MRI) define the type of mass by defining its extension and relationship with the adjacent anatomical structures. This helps in the development of an appropriate surgical plan that permits complete resection of the neoplasm, including coccygectomy. For this reason, these studies are crucial to the management of SCT [9].

The possibility of malignant transformation of SCT increases with age, with malignancy rates as high as 70% if SCT is diagnosed at one year of age. Serum alpha-fetoprotein (AFP) is frequently used as a tumor marker SCT and may be used during routine follow-up after SCT resection; however, the diagnostic ac-

curacy of serum AFP levels during follow-up has not been well established [10]. Fetal ultrasound can be used to diagnose SCT prior to delivery, and about 80% of patients receive a diagnosis within the first month of their lives [10]. Sacrococcygeal teratoma (SCT) has a tendency toward malignant degeneration, necessitating early surgery; the rate of recurrence following surgery has been estimated at 10% - 15%, the most significant risk factors for recurrence are incomplete resection and immature/malignant histology. Preoperatively, older age at diagnosis (>2 months) and predominantly solid components within the mass are suggestive of malignant histology and a poor prognosis, The removal of the coccyx bone and avoiding tumor tissue spillage during surgery have been stressed as ways to prevent incomplete resection [8].

4. Conclusions

Sacrococcygeal teratomas (SCTs) are uncommon congenital tumors that typically manifest in the neonatal period. As a child age, the likelihood of malignancy increases, highlighting the importance of early detection in lowering morbidity and mortality.

Although the diagnosis can be made with CT scan and MRI, anathomopathology studies can confirm it. The treatment is surgical, with the goal of achieving complete resection to prevent recurrence. Post-operative monitoring based on a neurological examination and urodynamic assessment is necessary to look for sphincter disorders. Rigorous follow-up with clinical examination, ultrasonography, and tumor markers is required to look for any recurrence.

Consent

Written informed consent was obtained from the patient parents for publication of this case report and accompanying images.

Author Contributions

All authors contributed to the conduct of this work. All authors also declare that they have read and approved the final version of the manuscript.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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