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Case Report

Adult sacrococcygeal teratoma with coccygectomy: A case report with a review of the literature

Daniel Diaz-Aguilar, Sergei Terterov, Rudi Scharnweber, Catherine Merna, Stephanie Wang, Shayan Rahman[1]

Department of Neurosurgery, David Geffen School of Medicine at UCLA, [1]Department of Neurosurgery, Kaiser Permanente Medical Center, Los Angeles, California, USA

E-mail: *Daniel Diaz-Aguilar - Ldiazaguilar@mednet.ucla.edu; Sergei Terterov - steterov@mednet.ucla.edu; Rudi Scharnweber - rscharnweber@mednet.ucla.edu; Catherine Merna - catmerna@gmail.com; Stephanie Wang - StephanieWang@mednet.ucla.edu; Shayan Rahman - Shayan.X.Rahman@kp.org
*Corresponding author

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Abstract

Background: Saccrococcygeal teratomas (SCT) are derived from embryonic germ cell layers. They frequently present at the base of the coccyx within the pelvis. While these tumors are common in children, they are exceedingly rare in adults. In adults, a majority of these tumors are intrapelvic and associated with a low risk of malignant transformation. Therefore, this contributes to a good prognosis following resection of mostly benign lesions.

Case Description: An adult female with chronic pelvic pain presented with a sacral teratoma. She failed conservative treatment and underwent a coccygectomy with an *en-bloc* excision of the tumor. Microscopic histological analysis showed no evidence of immature or malignant elements, confirming the diagnosis of a mature, benign, cystic SCT.

Conclusions: Mature SCTs in adults are rare malignant lesions. In this case, the patient was cured following primary surgical excision requiring *en-bloc* coccygectomy.

Key Words: Adult, mature teratoma, sacral coccygectomy

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INTRODUCTION

Sacrococcygeal teratomas (SCT) are multipotential cell tumors most commonly seen in neonates but only rarely in adults. SCTs carry a female preponderance of 4:1, and a prevalence of 1:30,000 births.[2] A vast majority of SCT are benign and have a low potential for malignancy. Although surgical resection carries an excellent prognosis, there is a high 37% postoperative recurrence rate for these lesions.[2,3] Here, we present the case of an adult female with chronic pelvic pain and an SCT, who was successfully treated with an *en-bloc* coccygectomy resulting in successful gross total removal of her sacral teratoma.

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CASE DESCRIPTION

Clinical presentation

A 23-year-old female presented with pelvic pain. Magnetic resonance imaging (MRI) of the pelvis revealed a complex cystic structure anterior to the coccyx [Figure 1]. The lesion, measuring approximately 3 × 2 × 4 cm, contained both cystic and proteinaceous components consistent with the diagnosis of a pre-sacral intraspinal teratoma; notably, it did not enhance with contrast. The differential diagnosis included meningocele and chordoma [Figure 2]. A fine needle biopsy anterior to the coccyx revealed scattered histiocytes and keratinocytes consistent with the diagnosis of an Altman type IV SCT. She failed conservative measures to treat her intractable pelvic pain, and ultimately underwent successful surgical intervention at the age of 26.

Surgery

The patient underwent an *en-bloc* posterior coccygectomy. Following exposure of the coccyx and lower part of the sacrum, the rectum was carefully dissected away from the tumor. An osteotome cut through the coccyx 6 cm above the tip [Figure 3]. The final tumor specimen was 1.9 × 2.5 × 1.6 cm. Gross analysis revealed an irregular tumor with cystic nodules attached to a firm portion of the coccyx [Figure 4]. The microscopic analysis confirmed a benign, mature sacrococcygeal cystic teratoma. The patient was discharged home without any complications. One year later, her she had no tumor recurrence or residual symptoms/signs.

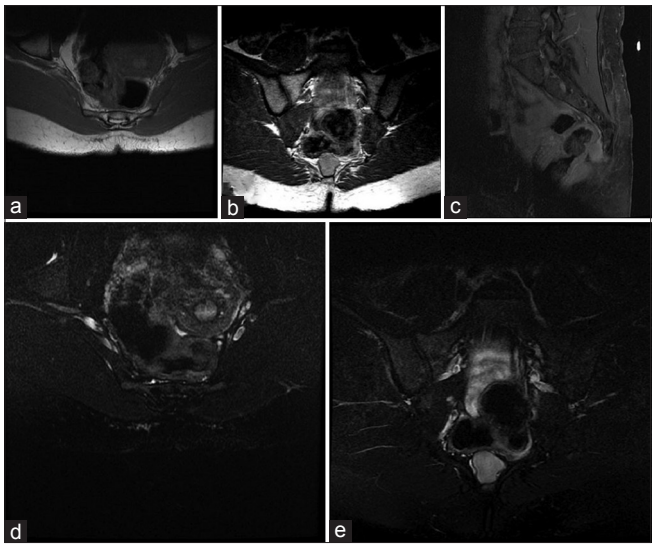

Figure 1: Axial T1-weighted (a), T2-weighted fat-saturated (b), sagittal T1-weighted post-gadolinium fat-saturated (c), coronal T1-weighted (d), and T2 fat-saturated (e) images demonstrating a complex precoccygeal cystic structure with multiple septations and fluid content of different signals and fluid levels

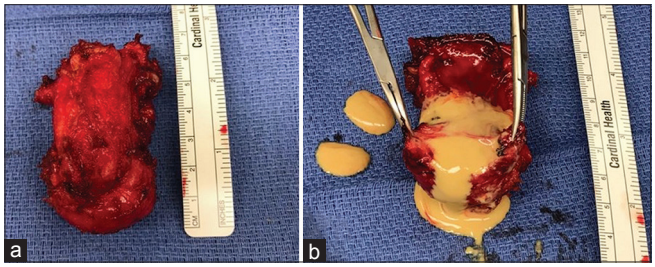

Figure 3: A solid cystic tumor (a, gross) containing yellow seromucoid fluid (b, gross)

DISCUSSION

Incidence and symptoms of sacrococcygeal teratoma

SCT are the most common fetal neoplasm. They account for 50% of teratomas in children but are rarely seen in adults.[3] The clinical presentation in adults includes bowel dysfunctions, urinary incontinence, lower back pain, and/or venous engorgement of the lower limbs.

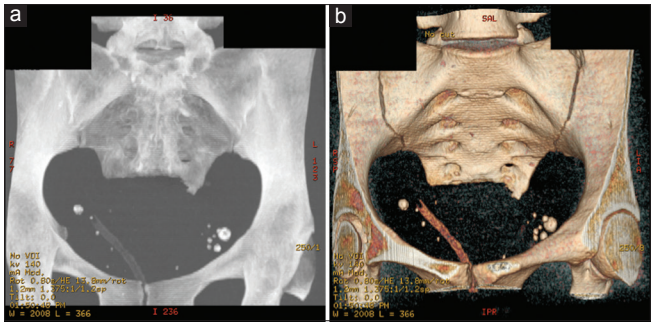

Figure 2: Coronal CT (a) and 3D reconstruction (b) of the coccyx status post resection of previously identified teratoma in the precoccygeal region

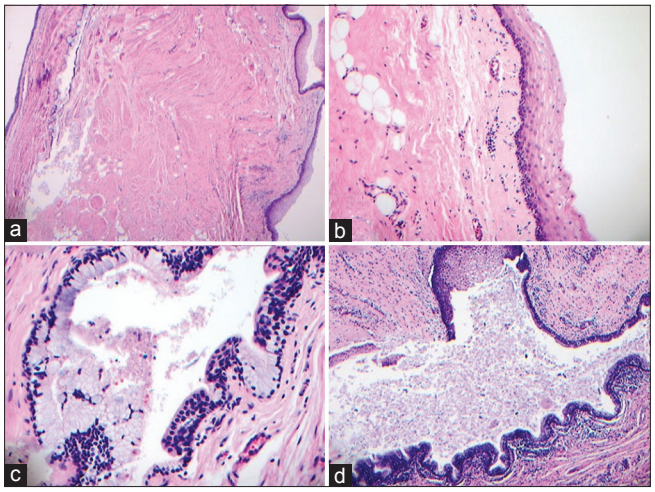

Figure 4: Histology of the mass demonstrating various types of mature tissue elements including lobules of smooth muscle, adipose tissue, stratified squamous epithelium (a, H and E, ×4; b, H and E, ×20) respiratory type ciliated epithelium (c, H and E, ×40; and mucous glandular tissue adjacent to stratified squamous epithelium (d, H and E, ×10)

Table 1: Classification of sacrococcygeal teratomas[3]

Altman type	Location	Incidence (%)	Malignant (%)
I	Entirely outside pelvis	46.5	8
II	Mostly outside pelvis	34.5	21
III	Mostly inside pelvis	8.75	34
IV	Entirely inside pelvis	12.75	38

Sacrococcygeal teratoma characteristics in adults

SCTs are divided into Types I–IV (criteria proposed by Altman *et al.*) [Table 1]. Most adult SCTs are type III and type IV; they are cystic and less liable to demonstrate malignant transformation (incidence 1–2%) compared to those seen in children and infants. Adult SCT are also primarily intrapelvic with no obvious physical cutaneous or dermatologic presence, and are often recognized following unresolved symptoms associated with a tumor mass compressing adjacent organs (e.g., vagina, uterus, or rectum).[4,5]

Histopathology and use of markers

Mature SCT can be distinguished from immature teratomas by the presence of epithelial structures along with well-distinguished cartilage and muscular tissue. Immature teratomas contain primitive combinations of germ layers with occasional mature elements.[1] The final diagnosis of mature vs. immature teratoma is dependent on histopathological examination. Several biochemical markers, including β-human chorionic gonadotropin (β-hCG) and α-fetoprotien (AFP), are used for initial screenings or the detection of tumor recurrences. However, their application is less applicable to mature teratomas as they often secrete fewer of these markers than immature or malignant teratomas.[2]

Predominance

Computed tomography and MRI are both vital preoperative diagnostic studies for the visualization of a SCT.[2] Our patient's studies revealed a lobulated solid and cystic mass in the presacral area anterior to the coccyx without calcifications.[5,7] These tumors frequently contain multiple cystic or solid components with necrotic areas that are highly suspicious for malignant transformation; however, the reported lesion was benign.[6] This SCT was curable with surgical excision/coccygectomy.

CONCLUSION

SCT, rarely observed in adults, are frequently benign, slowly growing intrapelvic masses best seen on MR studies. Mature benign SCT are highly susceptible to complete and early excision, without the additional need for chemotherapy or radiation.

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Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Al-Essa A-A, Malik TA, Baghdadi MK, Tayeb El AA. Adult sacrococcygeal teratomas. Saudi Med J 2004;25:367-9.
2. Allsopp G, Sgouros S, Barber P, Walsh AR. Spinal teratoma: Is there a place for adjuvant treatment? Two cases and a review of the literature. Br J Neurosurg 2000;14:482-8.
3. Altman RP, Randolph JG, Lilly JR. Sacrococcygeal teratoma: American Academy of Pediatrics Surgical Section Survey-1973. J Pediatr Surg 1974;9:389-98.
4. Audet IM, Goldhahn RT, Dent TL. Adult sacrococcygeal teratomas. Am Surg 2000;66:61-5.
5. Bruneton JN, Diard F, Drouillard JP, Sabatier JC, Tavernier JF. Primary retroperitoneal teratoma in adults: Presentation of two cases and review of the literature. Radiology 1980;134:613-6.
6. Miles RM, Stewart GS. Sacrococcygeal teratomas in adult. Ann Surg 1974;179:676-83.
7. Panageas E. General diagnosis case of the day. Primary retroperitoneal teratoma. AJR Am J Roentgenol 1991;156:1292-4.