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Adult Sacrococcygeal Teratoma: The third leg A rare case report in a 25 year old man

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ABSTRACT

INTRODUCTION: Sacrococcygeal Teratoma (SCT) is a rare benign neoplasm comprised of mixed elements derived from two or more germ cell layers. They are extremely rare in adults. They attract attention because of their gross appearance and bizarre histology.
PRESENTATION OF CASE: A 25 year old male presented to Surgical OPD, Government General Hospital, Guntur Medical College, Guntur with a large mass comprising a partially developed 3rd leg and rudimentary external genitalia in the lower back. He complained of cosmetic blemish, difficulty in sitting, sleeping and walking.
DISCUSSION: He was diagnosed mature Sacrococcygeal Teratoma Altman Type II based on history, clinical examination, ultrasound, MRI. Histopathology confirmed the diagnosis. He had complete surgical excision with primary wound closure and a good postoperative recovery. A 3-year follow-up using clinical, biochemical and radiological assessment revealed no evidence of recurrence.
CONCLUSION: Mature SCT, though very rare in adults, are usually benign. Complete surgical excision remains the mainstay of treatment.

1. Introduction

Sacrococcygeal Teratoma (SCT) is a rare tumor with predominant presentation in neonates with a prevalence of 1 in 40,000 births and a female preponderance of 4:1 [1]. They are attached to coccyx. They are believed to arise from totipotent somatic cells of the primitive knot [2], and thus, are composed multiple tissue types of 2 or 3 germ layers. Historically, teratomas are attributed to demons, sexual misconduct and abnormal fertilization [3]. Ultrasound allows prenatal detection of SCT from 2nd trimester.

SCTs are very rare in adults and as a rule are benign and have extremely low potential for malignancy probably due to their dormant nature [4].

The prognosis is excellent with prompt surgical excision including coccygectomy [5]. The complications of the mass effect and/or surgery may include neurogenic bladder, urinary, fecal incontinence, and other chronic problems resulting from accidental damage to/or sacrifice of nerves and muscles within the pelvis [6]. The most frequent complication is an unsatisfactory appearance of the surgical scar. Complications of not removing the coccyx may include recurrence (37%) and metastatic cancer [7].

2. Presentation of case

A 25-year-old male presented with a third leg in his lower back, which was noticed at birth and slowly grown to the present size. He complained of cosmetic blemish, difficulty in sitting, sleeping and walking. Bladder and bowel functions were normal. Grossly, it is a single non tender solid 35 cm × 20 cm × 10 cm mass extending between the mid-axial lines horizontally and from the highest point of the iliac crest to the tip of the coccyx with a short 4 cm leg and a well formed foot 17 cm × 8 cm with distinct heal, sole, dorsum and 5 toes overhanging the right buttock (Fig. 1). The limb showed good range of movements over its attachment to sacrococcygeal region. There were traces of rudimentary genitalia on the right side of the foot.

3. Discussion

X-ray revealed well-formed bones of foot, particularly long bones presumed to be femur and tibia. Plain MRI of lower lumbosacral region suggested L4/L5 block vertebral anomaly of sacrum with spina-bifida, prominent subcutaneous fat, few rudimentary bony structures, muscle elements and a well defined saccular

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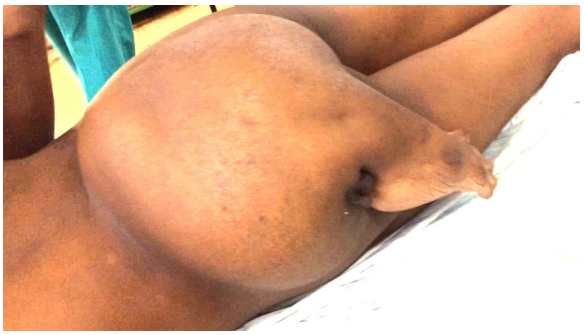

Fig. 1. Posterior view—showing mass extending from highest point of the iliac crest to the tip of coccyx with a short leg and well formed foot overhanging the right buttock.

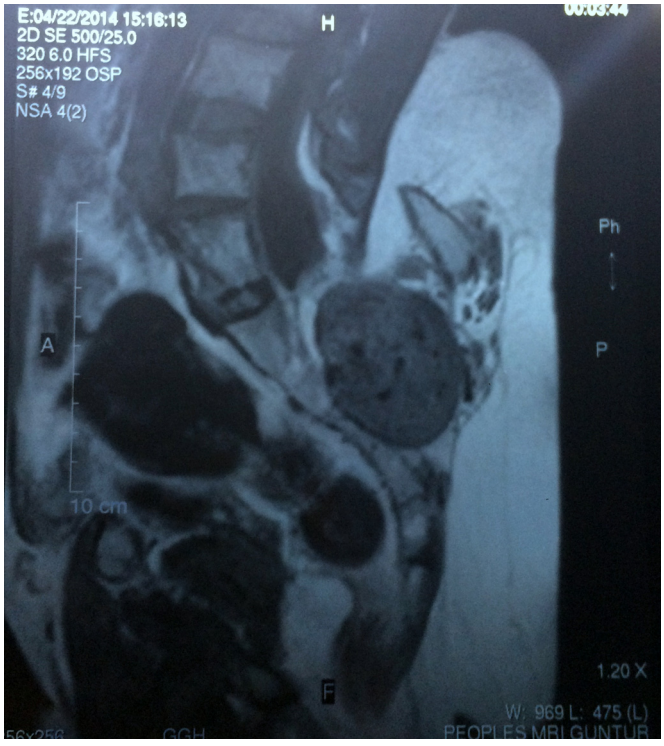

Fig. 2. Magnetic Resonance Imaging (MRI) scan—showing a mass in the sacrococcygeal area, L4/L5 sacral anomaly with spina bifida, subcutaneous fat, few bony structures, muscle elements and a bowel segment overlying the sacrum.

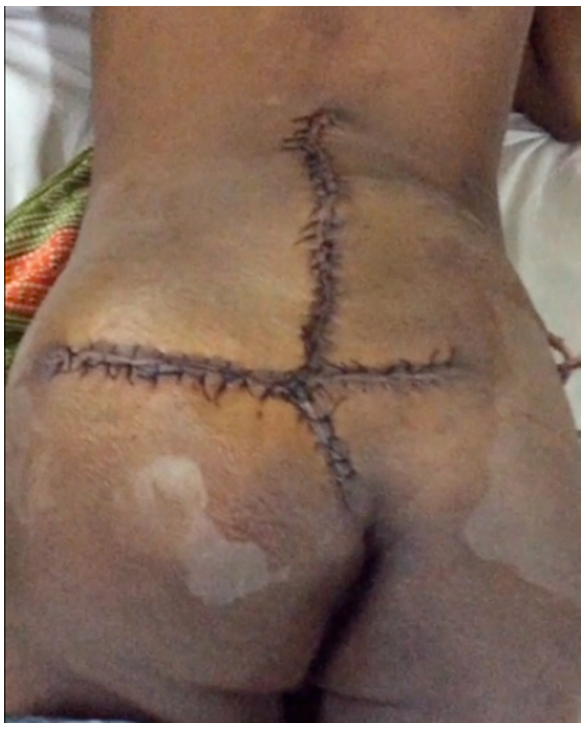

Fig. 3. Postoperative view of the patient—with SCT excised and flap reconstruction performed.

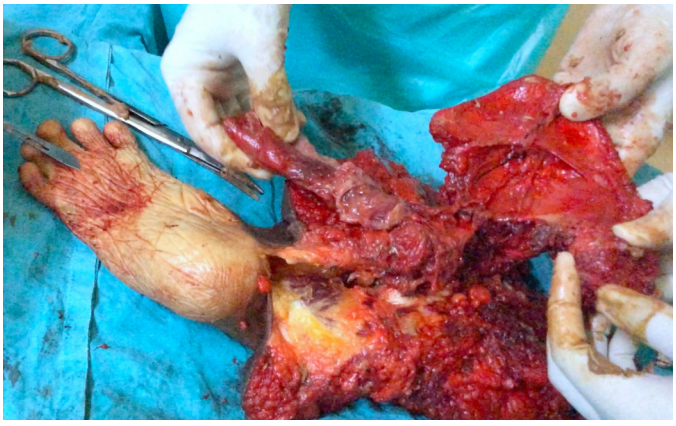

Fig. 4. Excised SCT specimen—with an accessory leg and a well formed foot, subcutaneous fat, muscle elements, bone and a bowel segment.

structure showing a pocket of air resembling bowel, overlying the sacrum (Fig. 2). There was no gross abnormality in pelvis. Ultrasound showed no other abnormalities. Alfa-feto protein levels were normal. The provisional diagnosis is Sacrococcygeal Teratoma-Altman type II (tumor mostly outside the body with small portion inside pelvis).

Excision of the SCT was planned under general anesthesia. A vertical elliptical incision was made encircling the mass and the limb. The limb along with the mass, muscles and bones were excised. Spina-bifida was identified and Dura was safely guarded. There is a triangular curved bone of size 8 × 6 cm present near the sacrum between which was a saccular cystic mass. This bony attachment was dissected with an osteotome and the mass was excised. Skin flap reconstruction was performed (Fig. 3). There were no intraoperative complications.

Histopathology revealed the tumor to be a mature teratoma with skin, subcutaneous fat, well differentiated bone resembling those of lower limb, rudimentary testis, bowel and lymphatic tissue (Fig. 4). There was no evidence of malignancy. Postoperative bowel and bladder functions were normal. Both legs showed normal motor and sensory functions. The healing was progressive with an acceptable scar. A 3-year subsequent follow up revealed no other complications.

4. Conclusion

SCTs account for 40% of all the teratomas in children and are extremely rare in adults. Till date, 7 cases of SCT with an extra limb are reported in literature. This is the 8th reported case of SCT with an extra limb and 3rd such case in adults [8]. Altman et al. reported the largest series of 400 cases of SCTs in 1974 and proposed the classification of SCT based on its extent [9]. The present case belongs to Altman type II.

SCT shows female preponderance (4:1) but our case is of a male. The reason for female preponderance is not known. As in this case,

Sacrum is the most common site of SCT. Modern ultrasound helps in prenatal detection [10], as this man is 21 years old, he was born in 1989, when the diagnostic techniques were not well established. His rural background, lack of serious complications despite of cosmetic blemish, unawareness is the reasons for the delayed presentation.

CT and MRI are beneficial to assess the blood supply and can further define the SCT's spatial relationship to surrounding organs [11]. The accessory blood supply in this case was found to be between the rectum and perirectal muscles. One needs to be vigilant with SCT and be cautious as the tumor could share a common blood supply with other vital organs. Surgical excision of the tumor with coccygectomy is the treatment of choice to prevent recurrences. In our case, the SCT was not attached to rectum or other important organs hence it allowed complete resection of the tumor once the main vascular and the accessory blood supply was controlled. This was combined with coccygectomy to avoid recurrences. The size of the tumor does not correlate with the likelihood of recurrence or poor outcome, but the age at diagnosis, treatment, histological evaluation and the stage at the time of resection are important prognostic factors.

Limb formation is seen more often but visceral organ tissue has also been reported. Miles reviewed 11 cases of SCT in adults with only one case of external presentation and the rest are intra pelvic presacral masses, which supports the unusual presentation in this case. Histopathology showed classical dermal derivatives skin, subcutaneous fat, well differentiated bone, rudimentary testis, bowel, and lymphatic tissue.

Follow-up during first three years of treatment is necessary to monitor recurrences and surgical complications. In the 3-year follow-up, he has maintained good urinary and ano-rectal function with no clinical biochemical and radiological evidence of recurrence. Longer period of follow up is required to make our claim more authentic.

Conflict of interest

None.

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None.

Ethical approval

Institutional ethics committee approval was obtained.
Ethics committee, Guntur Medical College, Guntur.
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Consent

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

Author contributions

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

Guarantor

Dr. Y. Kiran Kumar.

References

[1] R.M. Miles, G.S. Stewart, Sacrococcygeal teratomas in adults, Ann. Surg. 179 (1975) 676–683.
[2] K.M. Fadler, D.F. Askin, Sacrococcygeal teratoma in the newborn: a case study of prenatal management and clinical intervention, Neonatal Network 27 (2008) 185–191.
[3] H.G. Gatcombe, V. Assikis, D. Kooby, P.A. Johnstone, Primary retroperitoneal teratomas: a review of the literature, J. Surg. Oncol. 86 (2004) 107–113.
[4] R. Tuladhar, S.K. Patole, J.S. Whitehall, Sacrococcygeal teratoma in the perinatal period, Postgrad. Med. J. 76 (2000) 754–759.
[5] S.Y. Luk, Y.P. Tsang, T.S. Chan, T.F. Lee, K.C. Leung, Sacrococcygeal teratoma in adults: case report and literature review, Hong Kong Med. J. 17 (2001) 417–420.
[6] L.H. Holly, W.F. Alan, M.C. Timothy, J.H. Lori, P.J. Mark, W. Douglas, A. Scott, Sacrococcygeal teratoma: prenatal assessment, fetal intervention, and outcome, J. Pediatr. Surg. 39 (2004) 430–438.
[7] A. Zaccara, B.D. Iacobelli, O. Adorisio, M. Petrarca, R. Di Rosa, M.M. Pierro, P. Bagolan, Gait analysis in patients operated on for Sacrococcygeal teratoma, J. Pediatr. Surg. 39 (2004) 947–952.
[8] K. JItoko, Cama Sonal Nagra, Sacrococcygeal teratoma with an accessory limb an unusual presentation, J. Pediatr. Surg. 3 (2013) 42–44.
[9] R.P. Altman, J.G. Randolph, J.R. Lilly, Sacrococcygeal teratoma: American Academy of Paediatrics Surgical Section Survey—1973, J. Peaditr. Surg. 9 (1974) 389–398.
[10] S. Ravi, D.E. Nicholas, H. Juliet, Sacrococcygeal teratoma over two decades: birth prevalence, prenatal diagnosis and clinical outcomes, Prenatal Diagn. 28 (2008) 1048–1051.
[11] K.N. Ozoilo, M.G. Yilkudi, J.A. Ede, Sacrococcygeal teratoma in an adult female Nigerian, Ann. Afr. Med. 7 (2008) 149–150.