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Surgical resection of large sacrococcygeal teratoma in adults: Report of two cases

Taher Hawramy, Awder Khazendar, Seerwan Hasan, Mohsin Ahmad

ABSTRACT

Introduction: Here, we present two case reports of adult female patients with the pathologic diagnosis of large sacrococcygeal teratoma. Case Series: In the first patient (aged 19 years), pelvic ultrasonographic examination showed a large (85.9×88.7 mm) multiloculated complex cyst posterior to the cervix. No evidence of recurrence was observed post-resection. The histological examination of the specimen showed mature cystic teratoma with no malignancy. The second patient (aged 16 years) was with a history of slow progressive enlargement of a non-ulcerative mass in the gluteal region. Surgery was performed. On histopathological examination, the specimen showed chronically inflamed pilonidal sinus with mature cystic teratoma. Conclusion: These two cases suggest that we should put in mind the differential diagnosis of teratoma in sacrococcygeal region masses and it can be cured with surgery if the tumor mass was completely resected with the coccygectomy.

Keywords: Abdominal pain, Pelvic ultrasonography, Pilonidal sinus, Sacrococcygeal teratoma

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INTRODUCTION

Tumors arising in the presacral area are uncommon and account for about 1 in 10,000 hospital admissions [1]. Sacrococcygeal teratoma is one such uncommon tumor with an overall incidence of approximately 1 in 35,000–40000 live births [2–6] and a female to male ratio of 4:1 [3, 4]. However, in other studies [2, 7], the female to male ratio was found to be 10:1 with the incidence of sacrococcygeal teratoma [3, 7]. Most of the sacrococcygeal tumors are benign, and only 1–2% of the tumors are malignant [2].

In the present study, we report two cases of mature sacrococcygeal teratomas of Altman type IV with gross macroscopical removal by the sacrococcygeal (posterior sagittal) approach.

CASE SERIES

Case 1

History and Examination

A 19-year-old unmarried female presented with two attacks of right side abdominal pain within a week. The pain was associated with nausea, vomiting, frequent urination and feeling of heaviness in the perineum. She stayed at the hospital for one day each time she had the pain attack and was treated conservatively with intravenous fluid and simple painkillers, eight months prior to surgical intervention.

The patient had a history of a small bulge in the perineum on the right side and a small sinus just below her coccyx during her lifetime. She had no leg pain or paresthesias, no difficulties in urination or defecation, no weight loss, and was able to walk with no difficulties.

The patient had an uneventful medical and surgical history with a negative parental consanguinity and a negative family history for same. No irregularities were marked in the menstrual cycle since her menarche at 12 years. In the last two years, she had a decrease in the duration of bleeding (from seven to three days) but with a normal amount of flow.

On physical examination, lower limbs revealed normal muscle tone and power, intact sensation, normal reflexes and intact sphincters and rectal mucosa, which was further confirmed by digital rectal examination. A large cystic mass was observed pushing the rectum posteriorly with the mucous membrane moving over it.

Diagnostic evaluation

Pelvic ultrasonography showed a 85.9×88.7 mm multiloculated complex cyst posterior to the cervix (Figure 1). Pelvic magnetic resonance imaging (MRI) scan showed a large lobulated outline and hyperintense signal on T1- and T2-weighted images. No enhancement was seen after gadolinium contrast (Figure 2).

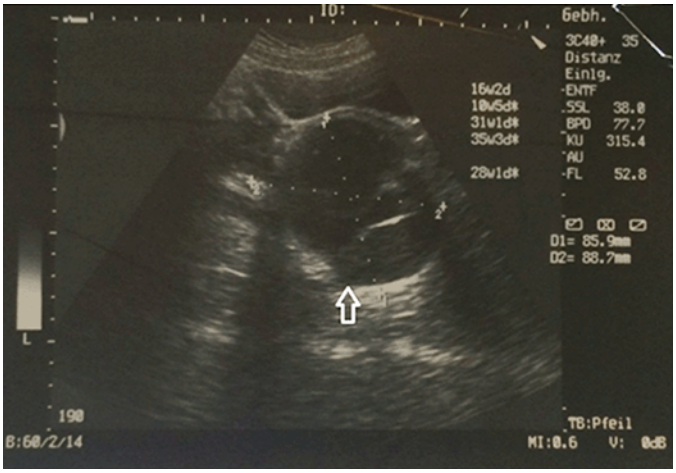

Figure 1: Pelvic ultrasonography of Case 1 showing the multiloculated complex cyst (arrow).

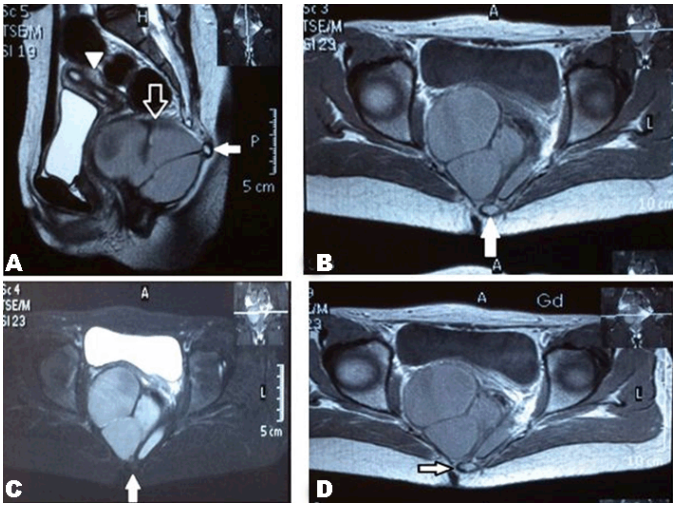

Figure 2: Magnetic resonance imaging (MRI) of Case 1 (A) T2-weighted images pelvic MRI scan (sagittal view) showing lobulated hyperintense mass (hollow arrow) anteroinferior to coccyx (solid arrow) and posteroinferior to uterus (arrowhead), (B) T1-weighted images of pelvic MRI scan (axial view) showing lobulated hyperintense mass close to coccyx (solid arrow), (C) T2-weighted images of pelvic MRI (axial view) showing lobulated hyperintense mass close to coccyx (solid arrow), (D) T1-weighted images of pelvic MRI scan with gadolinium contrast (axial view) showing non-enhancing lobulated hyperintense mass close to coccyx (outlined solid arrow).

Surgery

The patient was operated by the sacrococcygeal (posterior sagittal) approach in the knee-chest position. An elliptical incision was performed over the coccyx. A capsulated, gray-brown colored and multiloculated caseous material was found attached to the coccyx, which was subsequently removed (Figure 3).

The wound was packed with gauze flavored with iodine and left for secondary healing to take place with regular dressing till 21 days.

Histopathological examination

Two pieces of tissue sections were obtained. The smaller one was gray-brown and elliptical, and measured 6.5×3.5×1.5 cm, with the skin ellipse measuring 5.5×1.5 cm. The cut section was gray and contained a piece of bone and sinus measuring 3×2 cm. The other piece of tissue was brown, measuring 9.5×5.5×3 cm, loculated, and the cut-section contained multiple cysts.

Microscopically, the sections showed mature respiratory mucosa, squamous epithelium, mucinous gland and glial elements associated with fibrofatty tissue, bundles of nerve fibers and smooth muscle. It also contained coccygeal bone. The diagnosis was mature cystic teratoma.

Outcome

The patient remained free of pain and pelvic discomfort for four months with a normal level of tumor marker:

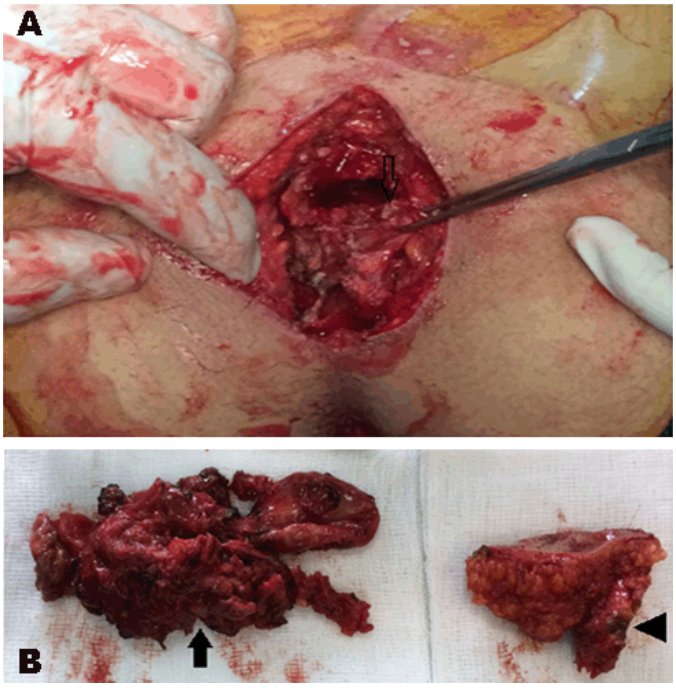

Figure 3: Intra-operative images of Case 1 (A) The capsulated multiloculated mass was attached to the coccyx which was subsequently removed; the cystic wall is grasped by the forceps (Hollow arrow), (B) The capsulated multiloculated gray-brown colored mass after removal; the left image is the cystic wall (solid arrow) and the right image is the skin with the pilonidal sinus (arrowhead).

alpha-fetoprotein 4.24 ng/ml (normal range: 0–5.9 ng/ml) and human chorionic gonadotropin <0.100 mlU/ml (normal range for non-pregnant female: <3.5 mlU/ml).

Magnetic resonance imaging of pelvis and lumbosacral region revealed it to be free of mass with the restoration of the position of the rectum, bladder and the site of coccygectomy (Figure 4).

Case 2

History and Examination

A 16-year-old unmarried female presented with progressive painful and an itchy mass on the right side of the gluteal region. This was red, tender, soft, fixed and a non-ulcerative mass. It was 1×1 cm when first noticed, but it progressively enlarged (10×7 cm) within three months.

The patient had a history of small sinus without discharge below the coccyx. She had no leg pain or paresthesias, no difficulties in urination or defecation, no weight loss and no walking difficulties.

The patient had uneventful medical and surgical history with a negative parental consanguinity and a negative family history for same. Her menstrual cycle was regular, with no change in the amount or duration. Her menarche started at 15 years.

Physical examination of lower limbs revealed normal muscle tone and power, intact sensation, normal reflexes, intact anal sphincter and normal rectal mucosa, which was

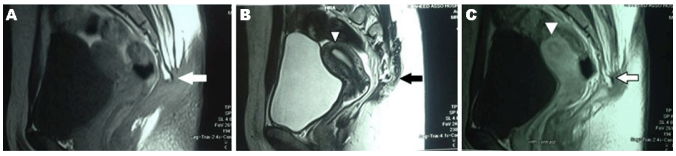

Figure 4: Postoperative magnetic resonance imaging (MRI) scan of Case 1 (A) T1-weighted sagittal view images of pelvic MRI scan showing no remnant of the mass with the site of coccygectomy (solid arrow), (B) T2-weighted sagittal view images of pelvic MRI scan showing no remnant of the mass with the site of coccygectomy (solid arrow) and restoration of the position of uterus (arrowhead). (C) T1-weighted sagittal view images of pelvic MRI scan with gadolinium contrast showing no remnant and enhancement of the mass with the site of coccygectomy (outlined solid arrow) and restoration of the position of uterus (arrowheads).

further confirmed by digital rectal examination. A cystic mass was found to be pushing on the rectum posteriorly; the mucous membrane was found moving over it.

Diagnostic evaluation

Pelvic ultrasonography showed a large cystic lesion (170×90 mm), extending from the right side of the coccygeal region to the posterior of the right adnexal region, and the cyst contained thick echogenic fluid.

Magnetic resonance imaging of pelvis and lumbosacral region revealed a large (17×15×10 cm) lobulated outline, hyperintense signal on T1- and T2- with a small (2×10 mm) fatty component near the coccyx, arising from the anterior of lower sacrum and extending posteriorly to the gluteal region more on the right side below the coccyx to the pelvis. It caused displacement of the rectum posterolaterally and uterus and urinary bladder anteriorly. No enhancement was seen after gadolinium contrast (Figures 5).

Surgery

The patient was operated under general anesthesia with endotracheal intubation in the prone position. An elliptical incision was performed over the coccyx which was subsequently removed. A cystic-like lesion containing paste-like material was found between the rectum and sacrococcygeal area. The lesion was separated from both the anterior surface of the sacrum and posterior wall of the rectum. After 48 hours, the wound was redressed with iodine flavored gauze and left open to be redressed twice daily till it had been closed after 28 days. There were no post-operative complications, and the patient was followed up for 1.5 years.

Histopathological examination

The tissue sections demonstrated a cystic mass lined by chronic inflammation composed of lymphocytes, xanthocytes, plasma cells and histocytes.

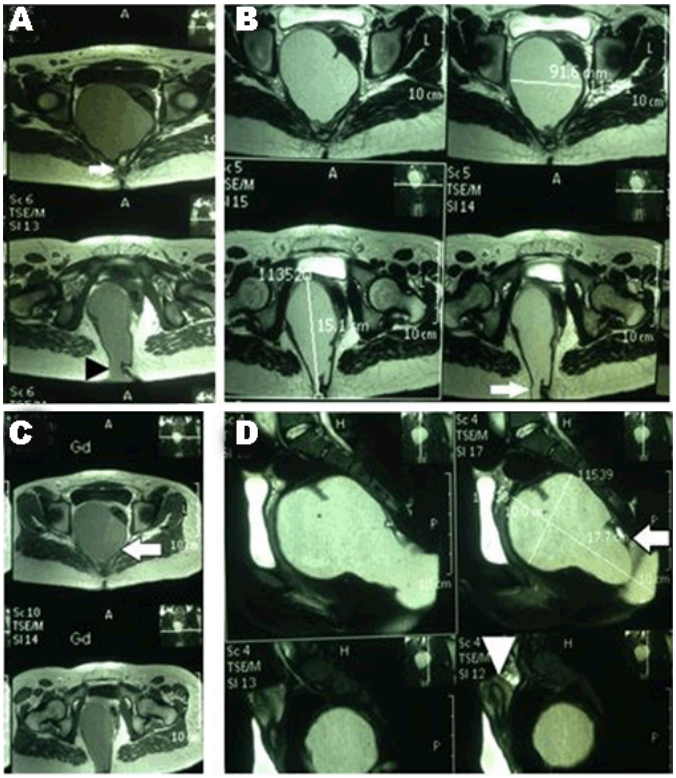

Figure 5: Magnetic resonance imaging (MRI) scan of Case 2 (A) T1 weighted axial view images of pelvic MRI scan showing lobulated hyperintense mass close to coccyx (solid arrow) and pilonidal sinus below coccyx (arrowhead). (B) T2-weighted axial view images of pelvic MRI scan showing the size of lobulated hyperintense mass with pilonidal sinus below coccyx (solid arrow). (C) Axial view pelvic MRI with gadolinium contrast showing no enhancement (arrow). (D) T2-weighted sagittal view images of pelvic MRI scan showing lobulated hyperintense mass anteroinferior to the sacrum and coccyx (solid arrow) and posteroinferior to the uterus (arrowhead).

A small cyst lined by ciliated epithelial cells was found on one side and squamous epithelial cells on the other, with enteric glandular component seen in the wall of the main cyst. A piece of the bone was present. The diagnosis was chronically inflamed pilonidal sinus with mature cystic teratoma.

Outcome

The patient is married and pregnant without any complications.

DISCUSSION

Sacrococcygeal teratoma is a congenital germ cell tumor, which is usually benign, and develops from the primitive knot or Hensen's node [1–3, 8, 9]. Sacrococcygeal teratoma can be classified according to its location (inside and outside the pelvis) as shown below [3, 8–10]:

- Altman type I: external with minimal pre-sacral involvement.
- Altman type II: external with significant extension into the pelvis.
- Altman type III: externally apparent but with a predominant pelvic mass that extends to the abdomen.
- Altman type IV: pre-sacral without external extensions.

Other studies [2, 9] classified it into mature (cystic or solid, benign), immature (malignant tissue of germ cell origin) and malignant (monodermal or highly specialized) tumors. The sacrococcygeal teratoma in both patients of this study was Altman type IV and both diagnosed histopathologically with mature cystic teratoma.

About 90% of the tumor presents with a visible external mass in neonates while most of the adult sacrococcygeal teratoma present as an intra-pelvic mass [2, 7]. Adult sacrococcygeal teratoma may be asymptomatic and discovered incidentally by digital rectal examination, vaginal examination or radiological investigations of the pelvis or peri-anal sinus. However, if the tumor is large enough to produce pressure effect, the patient may complain of pain in the perineum, constipation and bladder dysfunction, and there may be a rare occurrence of fistula in the bladder or rectum [1, 2, 9]. Neurological signs and symptoms are absent, and this makes it different from chordoma [1].

The investigations include abdominal and perineal ultrasonography, barium enema to know about bowel involvement and position, urography for the ureter position, arteriography to exclude tumor vascularity, CT scan, and MRI scan with the last two being the investigation of choice [1, 2]. We only performed pelvic ultrasonography and MRI for diagnosis.

Differential diagnosis includes sacral chordoma, lipoma, anterior myelomeningocele, rectal duplicated cyst and gland cyst, necrotic rectal leiomyosarcoma, pyogenic abscess, neurogenic cyst, cystic lymphangioma, neuroblastomas, dermal sinus stalks, retrorectal hamartomas and intracanalicular epidermoid tumors [2, 7, 8].

The approach to the mass may be trans-abdominal, trans-perineal, a combination of abdominal and perineal approach, combined abdominal sacral approach, or more recently laparoscopic approaches [2, 7, 9]. For low-lying presacral tumor by a vaginal approach has been reported in females [1]. We performed complete surgical excision with coccygectomy through sacrococcygeal (posterior sagittal) approach for both patients, with more than 95% cure rate depending on the malignancy [3, 9].

Postoperative complications with coccygectomy are delayed wound healing, infection, increase time for recovery and rare bowel herniation [9]. We did not face postoperative complications except for delayed wound healing.

CONCLUSION

Although sacrococcygeal teratoma is a rare case in adults, the present study considers teratoma as a differential diagnosis of the masses of sacrococcygeal area and it can be cured with complete surgical intervention, which shows extremely good prognosis.

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Author Contributions
Taher Hawramy – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Awder Khazendar – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Seerwan Hasan – Substantial contributions to conception and design, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Mohsin Ahmad – Substantial contributions to conception and design, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor
The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

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