

Case Report

A case report of large adult sacrococcygeal teratoma with literature review

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ABSTRACT

Sacrococcygeal teratomas rarely present in adulthood. Due to its rarity, accurate diagnosis is a clinical dilemma in adults. The patient was a 41-year-old female who presented with pelvic pain, low back pain radiating to the legs and chronic constipation. The patient underwent a CT scan which demonstrated a 6 cm presacral multiloculated mass compressing rectum. Prior to presentation at our clinic, she had an EUS and biopsy by gastroenterology, followed by diagnostic laparoscopy due to her past medical history of endometriosis. Her workup was negative for gynaecologic pathology which led to a surgical referral. We evaluated the images and took her to the operating room for posterior paracoccygeal retrorectal mass resection with coccygectomy and intraoperative flexible sigmoidoscopy. She had complete excision for pathologically proven mature 9 cm teratoma. Sacrococcygeal teratoma is rare in adults. A high index of suspicion is required for diagnosis. It should be in the differential diagnosis when dealing with presacral masses in adults. Complete excision is the treatment of choice.

Keywords: Sacrococcygeal teratoma, Endometriosis, Coccygectomy

INTRODUCTION

Sacrococcygeal teratoma is a rare childhood and infancy sarcoma.¹⁻³ Very few cases have been reported in adults.^{2,3,5,8} It is mostly cystic and benign. Due to the rare adult presentation, correct diagnosis is challenging and typically found incidentally.^{4,5} Management of teratoma is complete surgical excision to alleviate the symptoms and rule out malignancy.

CASE REPORT

A 41-year-old female presented to our office after a biopsy of presacral mass which was causing compression on her rectum. She has a medical history of migraines, gastroesophageal reflux disease, irritable bowel syndrome and endometriosis and past surgical history of cholecystectomy, tubal ligation and colonoscopy. Upon

presentation, her chief complaints were chronic pelvic pain, leg pain, worsening constipation and lower back pain for the last 3 months. She was worked up by her gynaecologist and gastroenterologist for her symptoms. A CT scan was done that showed complex cystic multiloculated 5.9×2.4×3.9 cm posterior lower rectal lesion extending to the sacral segment and coccyx compressing the rectum. She underwent colonoscopy and endorectal ultrasound which confirmed a cystic mass followed by FNA by GI. Pathology was significant for benign appearing squamous epithelial cells, few bland ductal cells and few inflammatory cells. To determine the origin of the lesion and suspected endometriosis, a diagnostic laparoscopy with biopsy and excision of an endometrial implant from the cul-de-sac was performed. Peritoneal fluid cytology was sent which showed atypical epithelioid cells, mesothelial cells, histiocytes and inflammatory cells inconclusive for malignancy. She was

referred to colorectal surgery for further management. Digital rectal exam confirmed an extraluminal palpable low posterior rectal mass with intact mobile rectal mucosa. No neurologic deficits or intergluteal fold skin abnormalities were noticed. She was taken to OR based on her prior workup. The perineal approach was chosen due to the tumor location below the S3 level. During surgery, it was confirmed to be a presacral mass below S3, multiloculated and involving the coccyx. The rectum was dissected off of the presacral fascia. She underwent posterior paracoccygeal retrorectal mass resection with coccygectomy and intraoperative flexible sigmoidoscopy. Her postoperative course was uneventful. The Final pathology showed an 8.8 cm mature teratoma. She was seen at 3 months follow up with resolution of her pelvic pain. She will have yearly follow-up and postoperative MRI evaluation.

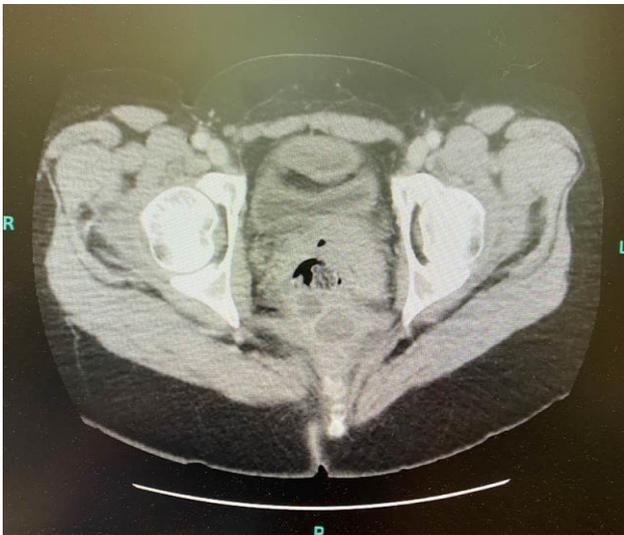


Figure 1: CT scan image showing complex cystic multiloculated 5.9X2.4X3.9 cm posterior rectal lesion extending to sacral segment and coccyx compressing rectum.



Figure 2: CT scan: Sagittal view of presacral cystic mass.



Figure 3: Coronal view on CT scan.



Figure 4: Sacrococcygeal teratoma after excision with dimensions.

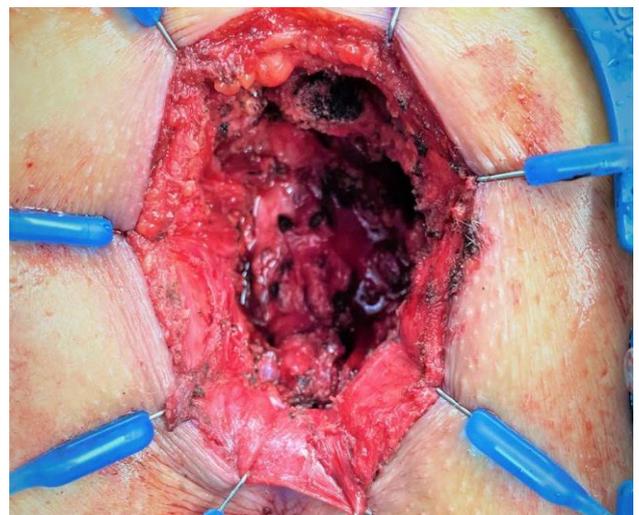


Figure 5: Intraoperative view after complete excision of sacrococcygeal teratoma.



Figure 6: Postoperative imaging with wound and JP drain in place.



Figure 7: Postoperative wound well healed sacral wound.

DISCUSSION

Teratomas are rare neoplasms. They originate from embryonic pluripotent cells and may have various degrees of maturation, according to which they are classified as mature, immature, and malignant. They may be inherently malignant or have the potential for malignant degeneration.

Sacrococcygeal teratomas are extremely rare. They are mostly present in infancy and childhood in this location, with an incidence of 1 in 40000 live births and a female to male ratio 4:1.^{1,2} Usually diagnosed prenatally, 50-70% present during the first few days of life, less than 10% are diagnosed beyond the age of two years. Malignant transformation has been found in approximately 1% of teratoma patients comprising squamous cell carcinoma, adenocarcinoma, sarcoma and other malignancies.

Adult presacral teratomas are rare tumors with an incidence between 1 in 40,000 and 63,000 and a 3:1

female preponderance. Fewer than one hundred cases are reported in the literature in adults.¹⁻³ The majority of adult presacral teratomas are cystic and benign. Only about 1-2% is malignant but there is a 20-30% risk of infection. Infants usually present with a visible mass, while adults present with pain and intrapelvic mass.^{3,4} Digital rectal exam or radiological imaging is common means of discovery and diagnosis. Symptoms may be subtle and nonspecific as a result of compression of adjacent structures: rectum, bladder or uterus. Common symptoms are lower back and pelvic pain, constipation, tenesmus, and frequent urination. If large enough, patients may complain of pain in the sacrococcygeal area, obstructive defecation, bladder dysfunction and neurological symptoms such as lower extremity numbness.^{4,5}

Differentiation between cystic changes in the ovary or presacral region is extremely difficult and a diagnostic challenge. Differential diagnoses for presacral cystic lesions are congenital abnormalities, cystic changes in the ovaries, meningocele, rectal duplication, tailgut cyst, neurogenic tumors, osseous lesions, renal cysts, Wilms' tumor, soft tissue tumors and metastatic tumors of the uterus or ovaries

Imaging studies should include transvaginal and transrectal ultrasound, computed tomography and magnetic resonance imaging, which are useful in identifying the exact location and characteristics of the tumor.⁶⁻⁸ These diagnostics are helpful for determining the optimal surgical procedure. Laboratory tests should include levels of AFP, β hCG, CEA, Ca-125, CA19-9 and CA 15-3, in case of suspicion of malignant transformation. Colonoscopy and sigmoidoscopy should also be considered.

Definitive diagnosis is possible only after histopathological examination. Therefore, the lesion should always be surgically removed. Drainage or biopsy is not recommended.

Complete surgical excision is the treatment of choice. Excision of the coccyx may be necessary because the bone may contain a nidus of pluripotent cells with a risk of recurrence.^{7,8} Surgical access to the tumor is possible via transabdominal approach or the transperineal route using the jack-knife position or a combined approach. Recently a laparoscopic-assisted approach has been described.^{9,10} If excision of the coccyx is considered the trans-sacral or the perineal route are more appropriate. Preoperative angiography may be utilized for embolization to reduce intraoperative hemorrhage, especially in large tumors. Complete excision has extremely good prognosis.

CONCLUSION

Adult sacrococcygeal teratoma is a rare entity. Mostly presents as benign presacral cystic mass with low

potential of malignant transformation. Clinical presentation is subtle. It is a diagnostic dilemma to differentiate between sacrococcygeal teratomas from other cystic pelvic masses especially ovarian on imaging. Definite diagnosis can be made only after surgical excision. Imaging is helpful in aiding diagnosis and for surgical planning. Complete surgical excision has extremely good prognosis and often times requires coccygectomy.

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REFERENCES

1. Afuwape OO, Ogundoyin OO, Ogunlana DI, Adeleye AO. Adult sacrococcygeal teratoma: a case report. Ghana Med J. 2009;43(1):40-2.
2. Valdiserri RO, Younis EJ. Sacrococcygeal teratomas: a review of 68 cases. Cancer. 1981;48:217-21.
3. Ghosh J, Eglinton T, Frizelle FA, Watson AJ. Presacral tumours in adults. Surgeon. 2007;5(1):31-8.
4. Mindaye ET, Kassahun M, Prager S, Tufa T. Adult case of giant sacrococcygeal teratoma: case report. BMC Surg. 2020;20:295.
5. Ng EW, Porcu P, Loehrer PJS. Sacrococcygeal teratoma in adults. Cancer. 2009;86:1198-202.
6. Gajula B, Srikanth K, Huda F, Sandhu H, Basu S. A Giant Sacrococcygeal Teratoma in an Adult Male: A Case Report. Cureus. 2011;13(3):e14181.
7. Mourad AP, De Robles MS, O'Toole S, Paver E, Winn RD. A case of an asymptomatic sacrococcygeal teratoma diagnosed in adulthood. J Surg Case Rep. 2020;2020(11):462.
8. Luk SY, Tsang YP, Chan TS, Lee TF, Leung KC. Sacrococcygeal teratoma in adults: case report and literature review. Hong Kong Med J. 2011;17:417-20.
9. Sukhadiya MV, Das U. Laparoscopic approach to type iv sacrococcygeal teratoma in an adult. Indian J Surg. 2015;77:62-3.
10. Cody HS, Marcove RC, Quan SH. Malignant retrorectal tumors: 28 years' experience at Memorial Sloan-Kettering Cancer Center. Dis Colon Rectum. 1981;24:501-6.

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