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Case Report

Asymptomatic adult type IV sacrococcygeal teratoma: incidental diagnosis in a case of subfertility

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ABSTRACT

Sacrococcygeal teratomas (SCT) are rare congenital tumours that are even more uncommon when present in adulthood. They are derived from residual stem cells in the presacral space that differentiate into clusters of somatic cells. We present a case of asymptomatic sacrococcygeal teratoma (SCT) in 28 years old female, presented for subfertility workup and diagnosed incidentally while investigating for it. Preoperative diagnosis was complex cystic lesion involving presacral space with possibility of dermoid, epidermal, or cystic presacral hamartoma. The mass was completely excised through abdomen and sacral approach. Histopathology confirmed it to be sacrococcygeal teratoma. Although rare, saccrococcygeal teratoma (SCT) should be considered in differential diagnosis of presacral masses in adult female. Complete surgical excision remains the cornerstone of treatment with the involvement of multidisciplinary approach.

Keywords: Sacrococcygeal teratoma (SCT), Magnetic resonance imaging (MRI), Pouch of douglas (POD)

INTRODUCTION

Sacrococcygeal teratomas (SCTs) are most commonly present in infants, and are exceptionally rare in adulthood. By definition, these germ cell tumours originate from pluripotent cells in the presacral space that differentiate into clusters of cells from any of the three primitive cell layers.[1] The estimated incidence is 1:40 000 with a 4:1 female preponderance. In adults, these tumours are nearly always entirely intrapelvic with no external component.[2] Adults cases often represent tumours that were present at birth (congenital), but not detected until adulthood. Most of these represent slow growing tumours that originated prenatally. Here, we present a case where the diagnosis was uncovered incidentally in an adult female presenting to our unit for subfertility.

CASE REPORT

A 28 years old housewife, nulligravida, presented to gynaecological outpatient department with inability to conceive since, 2 years. No history of pressure symptoms, coital difficulty, contraception uses or male factor abnormality noted. She had a history of laparotomy six months back but cyst could not be found so abdomen was closed. She was given ovulation induction for 2 cycles post operatively, but no response so, she was referred to Indraprastha Apollo Hospitals, New Delhi. Per abdomen findings showed tenderness in lower abdomen. Per speculum examination showed bulge in post vaginal wall simulating rectocele but firm in consistency, 2 cm from introital opening and extending to presacral area. Cervix could not be visualised. Per vaginal examination showed firm mass in post vagina extending up to presacral area pushing uterus and cervix anteriorly high. Digital rectal examination showed firm mass towards right of rectum.

Lab investigations revealed normal blood counts and including liver and renal function test. Serum tumour marker Ca 125 and Ca 19/9, were raised, 190.5 and 186.9 respectively rest others tumour markers (HCG, AFP, LDH) were normal. USG lower abdomen, done six months

back showed anteverted normal size uterus with right mutliloculated ovarian cyst 32×16 mm, 30×20 mm. A well-defined heterogeneous area with internal echoes 96×95×80 mm vol (385 ml) seen in POD and right adnexal region abutting/inseparable from right ovary? dermoid and endometriotic cyst. Repeat ultrasound done after 6 months showed anteverted normal uterus, RO with 2, 19.7 mm and 19.9 mm follicles, 125×105×97 mm size heteroechoeic (predominantly hyperechoic with anechoiec areas within) well defined area seen posterior to uterine midline / right paramedian region abutting of ovary? dermoid. No evidence of free fluid in pelvis/POD.

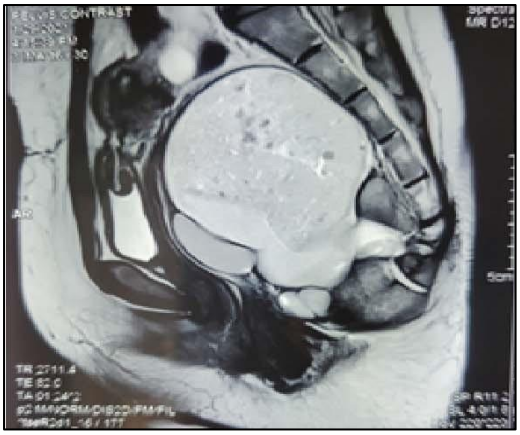

Figure 1: Pelvic contrast MRI revealed multiloculated well defined multiseptated thin walled T1 hypointense and T2 hyperintense lesion of size 133×90×96 mm noted within presacral space with effacement of adjacent mesorectal fascia. T2 signal intensity of different loculi are variable in signal intensity with evidence of mild multifocal T1 and T2 dark nodules within few septae.

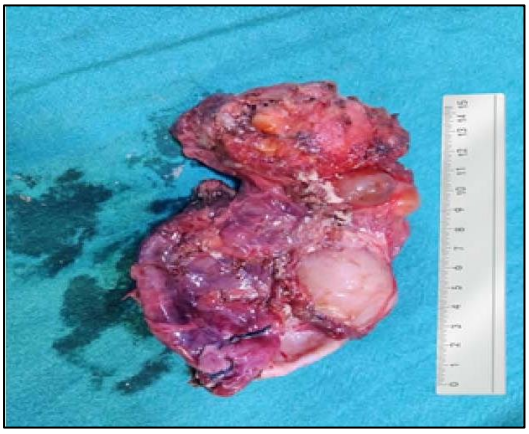

Figure 2: Shows solid cystic tumor mass with multiple cystic locules filled with thick whitish mucoid secretions, yellowish areas of fatty tissues, cheesy material and hairs noted at places.

Pelvic contrast MRI revealed multiloculated well-defined multiseptated thin-walled lesion 133×90×96 mm noted within presacral space with effacement of adjacent mesorectal fascia. Anorectal canal mildly displaced anterolateral on left side. Findings consistent with likely complex cystic lesion involving presacral space with possibility of dermoid/epidermoid or cystic presacral hamartoma 31×27 mm right ovarian cyst with haemorrhage within likely periovulatory right ovarian cyst.

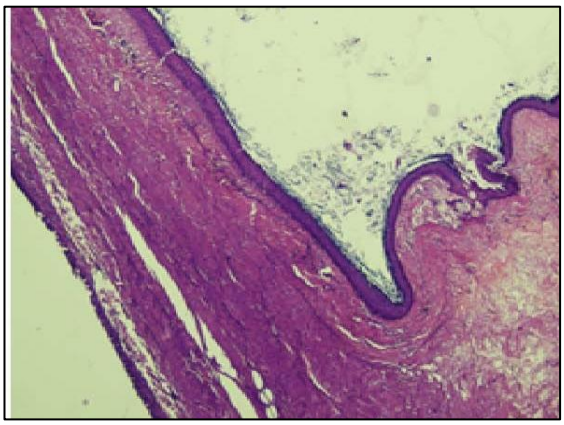

Figure 3: HE stains (4X magnification) showing mutiloculated cystic neoplasm lined variably by squamous epithelium and respiratory type ciliated epithelium, which overlies dense fibrocollagenous wall.

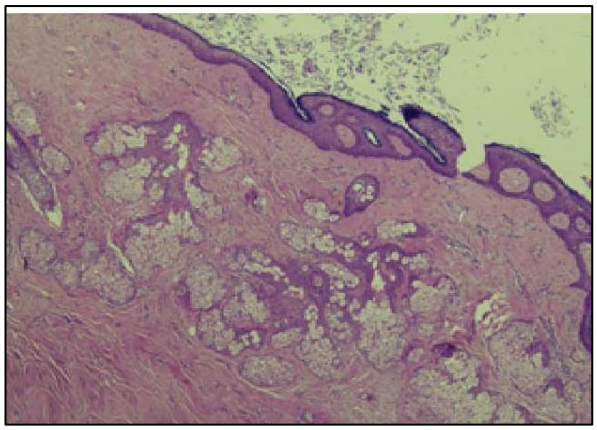

Figure 4: HE stains (4X magnification) showing cystic wall with lobules of sebaceous glands and hair follicles.

After taking consent and crossmatching 2 unit of PRC, with the involvement of multidisciplinary team she was taken for laparotomy with prophylactic DJ Stenting, right ovarian cystectomy and removal of presacral mass. Controlled needle aspiration of mass showed cheesy sebaceous material with hairs coming out of mass, which confirmed its benign nature. Decompression helped us dissect it off presacral fascia and rectum in the narrow pelvis. Abdominal and perineal approach was adopted with patient in the Lyod Davis position and a midline abdominal and perineal incision was made between ano coccygeal region. Since abdomen was open, it was

difficult to keep the patient in prone position. The muscles were retracted for access into the presacral space where tumour was bulging out in the perineum and ischiorectal fossa and the lobulated cyst was dissected off the presacral fascia, anococcygeal ligament posteriorly and anal sphincter complex anteriorly using ligasure and the specimen was retrieved through the perineum. Anorectal sphincter integrity checked. Abdominal and perineal drain kept for drainage. Post operatively, she recovered well with no residual bladder or bowel dysfunction, tolerating normal feeds. Patient discharged with perineal drain which was removed after 2 weeks.

Histopathology confirmed it to be saccrococcygeal mature cystic teratoma (SCT). Sections from the mass in retroperitoneum revealed a benign solid cystic neoplasm with several cystic locules. The smaller thin-walled cystic locules are lined variably by enteric type tall mucinous columnar epithelium, attenuated mucinous columnar epithelium, respiratory type epithelium and focally by urothelium. The tissues are represented by hair follicles, sebaceous glands, eccrine glands, few melanin pigment-laden nevus cells, lobules of mature adipocytes, few small plates of lamellar bone and cartilage, and smooth muscle bundles. There is patchy lymphocytic infiltrate in the cyst wall. No Immature tissues or primitive neuroepithelium and evidence of malignancy seen.

DISCUSSION

Adult SCT is mostly seen as an intra-pelvic mass when compared to the neonatal variety which mostly presents as externally visible mass.[1] The sacrococcygeal region is the most common site for a teratoma in infants. Sacrococcygeal teratomas affecting adults is extremely rare.[3] Most adult SCTs are cystic, and 1 to 2% are malignant. They are derived from Hensen's node; a focus of pluripotent stem cells that migrate caudally in the developing embryo and reside anterior to the primitive coccyx.[4]

Sacrococcygeal teratomas are classified according to the American academy of pediatrics surgery section survey into four types based on the amount of mass present externally versus internally.[7]

Type I-Sacrococcygeal teratoma, the mass is external with minimal or no internal components. Type II-Predominantly an external mass with internal extension into the presacral space. Type III-External and internal both. Type IV-Entirely internal with no external component.

The clinical presentation of SCT depends on its location and extension. Incidental features have been reported during routine clinical assessment. Symptoms may be related to mass effects of the tumour, such as low back pain, bowel or urinary symptoms, and venous engorgement of the lower limbs.[2] Patients may also present with an abscess or a perirectal fistula. Extrinsic compression of the vagina, as well as displacement of the uterus and rectum, may also be detected on pelvic and rectal examination as seen in our case. In rare cases, sacrococcygeal tumours cause partial paralysis (paresis) of the legs and tingling or numbness (paresthesia).

In adults, a diagnosis of sacrococcygeal teratoma may be suspected during a routine pelvic or rectal examination that detects the presence of a mass or tumor. MRI has superior specificity and accuracy than CT to visualize the soft-tissue extent in SCT.[4,10] Elevation of serum tumor markers such as alpha-feto protein (AFP), carcinoembryonic antigens (CEA), human chorionic gonadotropin (HCG) and lactate dehydrogenase (LDH) are usually suggestive of malignant transformation. Differential diagnoses of a presacral mass include an inflammatory abscess, tailgut cyst, metastatic deposit or lesions of neurological origin such as a meningocele.[3,6]

Surgical excision is indicated for SCTs that are symptomatic, suspected to be malignant and in women with child-bearing potential.[6] The latter is particularly important due to the potential for obstetric complications during vaginal delivery.[8] The prognosis of benign SCTs following complete resection is excellent, although there is a small lifetime risk of local recurrence. Malignant lesions carry a poorer prognosis and surgery is usually followed by adjuvant therapy tailored to the tissue histology.[9] Removal of the coccyx lowers the chance of recurrence. Chemotherapy and radiotherapy in addition to surgery may be indicated for malignant lesions

CONCLUSION

Although a rare tumour in adults, SCT should be considered in the differential diagnosis of patients with occult pelvic masses. For an occult pelvic mass not visualised by laparotomy/laparoscopy, cross sectional imaging CT and /or MRI is advocated for better anatomical delineation. Complete surgical excision remains the mainstay of treatment. A multidisciplinary approach is involved in the management of such cases, which presented as subfertility.

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