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Sacrococcygeal teratoma: Case report and review of literature

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Abstract
Sacrococcygeal teratoma is a group of rare congenital tumors. Arising from germ cells, they are predominantly seen among females. Usually the diagnosis is made antenatally through Ultrasonography. In adults they are usually asymptomatic and may be found incidentally. These are mostly benign but can transform into malignant lesions. Complete surgical excision is the mainstay of treatment and both open and laparoscopic techniques have been shown to be effective. We present such rare case of presacral type teratoma in young female

Keywords: Sacrococcygeal teratoma, teratoma, congenital tumor, sacral mass

Introduction
Sacrococcygeal teratomas (SCT) are a group of congenital tumors with a reported incidence of about 1/35000 to 1/40000 live births [1]. It has been shown to be predominantly associated with female sex with a female: male ratio of 4:1 [2]. It is commonly diagnosed in antenatal period or in neonatal period and is very rare finding in adults. Most of these tumors are benign in nature with a malignancy rate of 1%. However, risk of malignancy increases with age [3]. Due to rarity of such cases, there is limited evidence available in literature regarding management. Here we present a rare case of presacral SCT in a 17-year-old female.

Case Summary
17-year-old female presented to surgery OPD with complaints of intermittent discharge from natal cleft since birth. It was serous in nature, clear in appearance and thin in consistency. There was no history of pain over the natal cleft and no history of fever. She did not have any bowel or bladder complaints. There were no neurological symptoms over bilateral lower limbs
On Examination there were two discharging sinuses (~2mm) one over natal cleft and one at left buttock. There were no local signs of inflammation but a tuft of hair could be seen projecting from the sinus.
Examination findings were pointing towards a diagnosis of pilonidal sinus except the complaints were since birth.
A contrast MR fistulogram was requested which showed a large, round retrorectal mass, with predominant fatty content with few peripherally enhancing loculated cystic areas and a fistulous tract to skin in left gluteal region suggestive of Sacrococcygeal teratoma (presacral type).

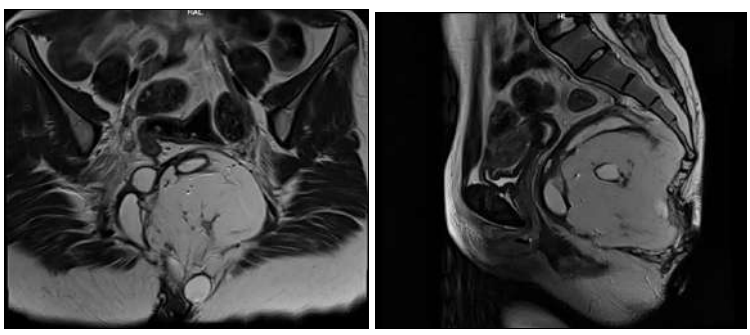

Fig 1: MRI showing large, round retro-rectal mass with predominantly fatty content

Elective resection was planned through an anterior abdominal approach. Intra operatively we noted a 12 x 10 x 10 cm mass adhered to posterior wall of rectum anteriorly and sacrum posteriorly. It was heterogenous soft to cystic in consistency and contained predominantly fibro fatty tissue with cystic areas containing hair.

Post op histopathology showed presence of cystic areas lined by stratified squamous epithelium with presence of keratinization(A) and pilosebaceous units(B) and inflammatory cell infiltrate comprising of lymphocytes (C), plasma cells, foamy macrophages. Heterogenous areas containing cartilage (D), skeletal muscles, neural tissue were also seen. A diagnosis of mature cystic teratoma was made.

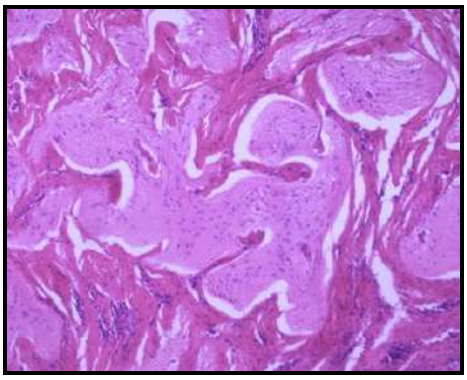

Fig A: epithelium with presence of keratinization

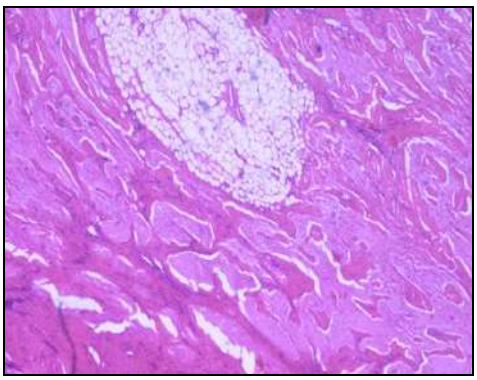

Fig B: Pilosebaceous units

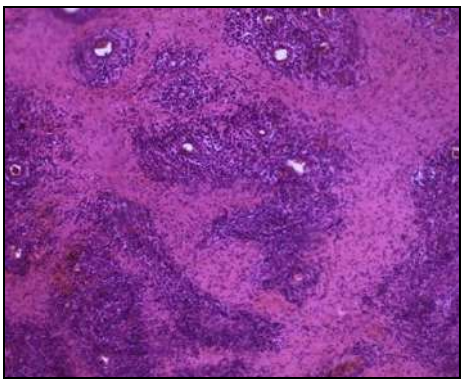

Fig C: Plasma cells, foamy macrophages

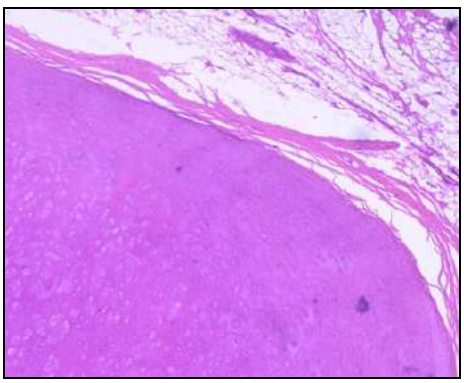

Fig D: Heterogenous areas containing cartilage

Discussion

Most common sites of teratomas are ovaries and testes of adolescent and these are thought to arise from germ-cells Teratomas can also be found in midline structures such as sacrococcygeal presacral, retroperitoneum postanal or even anterior mediastinum and pineal gland [4]. Tumors originating in sacrococcygeal region may be found ventral or dorsal to the sacrum and they may grow posteroinferiorly into the gluteal region or anterosuperiorly into the pelvis. Especially in the ventral location these tumours may grow to a large size as they develop into the retrorectal or presacral space [5].

Altman *et al*. classified sacrococcygeal teratomas in 4 types on the basis of their location with respect to the sacrum. Type I tumors are predominantly external with minimal presacral component. Type II tumors present externally but with significant intrapelvic extension. Type III is still apparent externally but predominantly a pelvic mass extending into the abdomen. Type IV is a presacral mass with no external presentation [6]. Our case conform to type IV of Altman's classification, although type III is most common seen in adults.

SCT are usually asymptomatic in adults and detected as incidental findings on imaging studies. Sometimes they may present with nonspecific symptoms or symptoms due to compression of adjacent structures such as rectum, bladder and uterus [7]. Symptoms may range from mild nonspecific abdominopelvic pain, back pain, constipation, urinary frequency, or dysmenorrhoea. If the growth is present externally, overlying skin may show discoloration and puckering [8].

Imaging studies form the backbone of investigative workup. CT and MRI are the most significant tools to characterize the mass, to evaluate the intrapelvic extension and relationship to adjacent structures. MRI has been shown to be superior to CT in terms of specificity and accuracy for visualizing the soft-tissue extent in SCT, containing abundant fat and fluid [9]. Since these tumors arise from germ cells, serum tumor markers (AFP, HCG, LDH) should be done and any elevation may be suggestive of malignant transformation [10].

The differential diagnosis of SCT in adults includes chordoma, meningocele, giant cell tumor of sacrum, osteomyelitis of sacrum, pilonidal cysts, rectal duplication cysts, fistula with presacral extension and abscess formation, post injection granuloma, and tuberculosis [11].

Complete surgical excision is the mainstay definitive treatment of SCT [12]. Both laproscopic and open procedures have been shown to be equally effective [13]. Coccyx is also excised to reduce the risk of tumor recurrence, which is reportedly 30-40% without coccygectomy. Such high recurrence rates are though t to be due to presence of nidus of totipotent cells in the coccyx

For tumors with a benign histology, complete excision is adequate. For malignant teratomas, surgical excision alone is inadequate, and patients should receive adjuvant chemotherapy and/or radiotherapy. But due to rarity of these tumors, there has

been no standard recommendation for the use of chemotherapy or radiation.

Acknowledgement: Nil

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