CASE REPORT

Presacral Epidermoid Cyst in an Elderly Female, Mistaken for Ovarian Cystadenoma

Jha Abhishek · Khalid Mohd · Gupta Prakhar ·
Saud Mohd · Gupta Gagan

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Introduction

Epidermoid cyst is of common occurrence in other parts of the body, but has rarely been described in the presacral space [1]. It is a benign congenital lesion, mostly seen in middle-aged females and is usually asymptomatic. Pathologically, it contains a thin wall lined by stratified squamous epithelium with a mixture of desquamated debris, keratin, cholesterol, and water. From the development aspect, it arises from the sequestration of the ectoderm following the closure of the neurenteric canal, proctodeal membrane, or post-anal gut [2]. The major complications of epidermoid cysts include infection and fistulization, where they may cause rectal pain.

Case Report

A 60-year-old woman presented to the gynecology OPD with the chief complaints of chronic constipation, tenesmus, and right flank pain. The general examination of the patient was

uneventful, except for pallor. The digital rectal examination of the patient revealed a soft mass in the right perirectal region. She was referred to the department of radiodiagnosis for further evaluation. Her transvaginal ultrasonography revealed a well-defined unilocular cystic mass in the pelvis with no internal calcifications, predominantly in the pouch of Douglas (Fig. 1). The right ovary could not be visualized. Based upon the clinical and sonographic findings, a provisional diagnosis of ovarian cystadenoma was made.

To accurately delineate the extent of the mass lesion and to rule out other pathologies, the patient was subjected to contrast-enhanced CT scan which revealed a well-defined fluid density and unilocular cystic mass with mildly enhancing wall anterior to the sacrum along the right lateral aspect of the rectum. The wall of the lesion contained few fine punctate calcifications (Figs. 2, 3). No evidence of destruction of the sacrum was seen. There was no evidence of regional lymphadenopathy. The rectum appeared to be compressed toward the left, and the urinary bladder and the uterus were displaced anteriorly. The fat planes with adjacent organs appeared well maintained. Based upon the imaging findings, the possibility of a retroperitoneal dermoid and epidermoid was maintained. A key feature that ruled out the possibility of adnexal origin was the location of the mass as adnexal masses usually do not extend in the presacral space. Moreover, hematological tumor markers of ovarian neoplasm including serum CA-125, AFP, and β -hCG were within normal limits.

The patient underwent laparotomy through the anterior transabdominal approach. After the midline incision was

Jha A. (✉) · Khalid M. · Saud M. · Gupta G.
Department of Radiodiagnosis, Jawaharlal Nehru Medical
College, Aligarh Muslim University, Aligarh,
Uttar Pradesh, India
e-mail: abhi.jnmc@gmail.com

Gupta P.
Department of General Surgery, Jawaharlal Nehru Medical College,
Aligarh Muslim University, Aligarh, Uttar Pradesh, India

made, a well-defined cystic lesion was seen along the right lateral aspect of the rectum, posterior to the uterus. There were no adhesions and the ovaries and fallopian tubes were normal. The cyst was resected, which measured 8×10 cm. The lesion was sent for histopathologic examination which confirmed an epidermoid cyst with high protein content (Fig. 4). The post-operative course of the patient was uneventful and she was discharged in a satisfactory condition 14 days post-surgery. She has been in regular follow-up for 1 year and her clinical assessment and routine abdominal sonographic examinations show no evidence of any residual lesion or recurrence.

Discussion

Presacral cystic masses are extremely rare with an incidence of 1 in 30,000–43,000 live births [3]. According to the estimates of the Mayo clinic, these lesions account for 1 in 40,000 hospital admissions [4]. A majority of these lesions are congenital, which include nearly 55–65 % of these lesions and predominantly include developmental cysts. These presacral cystic lesions include epidermoid cysts, dermoid cysts, neurenteric cysts, and tailgut cysts. Epidermoids are some of the rarest lesions among them. In a retrospective study done by Yang et al. [5], presacral epidermoids were the second most uncommon presacral masses.

Epidermoid cysts are congenital benign cystic lesions that are derived from the ectoderm. Characterization of various presacral cystic masses on imaging is difficult. The clinical presentation is varied, though a majority of the patients are asymptomatic. Most of the symptoms are attributed to the mass effect of the cyst, which is governed by the cyst size. The most common complaints of the patient include constipation, pain and discomfort during defecation, and vague lower abdominal pain and hydro-nephrosis due to the mass effect [6]. Many of the cysts become symptomatic after superadded infection, which can result in chronic non-healing sinus, perineal fistulas, or bleeding per rectum [7, 8]. These cysts usually affect middle-aged and elderly females, though the occurrence of presacral epidermoid has even been reported in a 4-year-old child [9, 10]. The major complications of these cysts include infections and bleeding. Malignant transformation is a rare but dreaded complication [11].

Although the radiologic literature regarding presacral epidermoids is sparse, certain features can help differentiate it from other lesions. Barium enema may show extrinsic rectal compression or widening of the retrorectal space on lateral images, but these findings are non-specific. Ultrasonography usually shows a cystic mass with or without internal echoes. CT scan reveals a unilocular or

multilocular presacral retroperitoneal mass [9], which on non-contrast scans may appear hyperdense owing to the high protein content or prior hemorrhage. In the case of superadded infections, the cyst wall may be thickened with abnormal enhancement; in the case of fistulization, air may be detected within the cyst. The MR imaging features of these cysts are similar to those of intracranial epidermoids. They appear heterogeneous on both T1W and T2W sequences owing to the keratin and the cholesterol. Most of the developmental cystic lesions of the presacral space exhibit homogeneous signal intensity on MR imaging. Though infection, hemorrhage, or calcifications in these lesions may alter the signal intensities, subtle changes in signal intensity favor epidermoid cysts.

The common differentials of retrorectal cystic lesions include tailgut cysts, cystic teratomas, neurenteric cysts, and anterior sacral meningoceles. The absence of fat may help

Fig. 1 Transvaginal scan showing a well-defined unilocular mass with internal echoes and few internal septations in presacral space with no internal vascularity

Fig. 2 Contrast-enhanced axial CT scan showing well-defined fluid density and unilocular cystic mass with mildly enhancing wall containing few fine punctate calcifications (White arrow)

Fig. 3 CT Scan at a caudal level reveals few fine septa within the mass (White arrow)

Fig. 4 Haematoxylin- and eosin-stained section shows cyst wall consisting of stratified squamous epithelium and laminated layers of keratin toward the lumen

in differentiating it from teratomas, and a well-defined smooth margin with the absence of invasion may help to differentiate it from malignant masses. Anterior meningocele is usually associated with sacral bony defects. However, none of these imaging findings are specific and so further workup is necessary.

Though many different surgical procedures like omentopexy have been tried in the management of these lesions [12], the ideal treatment for presacral epidermoids is complete surgical excision with complete removal of the epithelial lining of the cyst to prevent complications, particularly malignant degeneration.

Conflict of interest The authors report no conflicts of interest.

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