A Rare Coexistence of Retrorectal and Ovarian Cysts: A Case Report

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Abstract

Introduction: Retrorectal cysts are rare benign lesions which are frequently diagnosed in middle-aged females. According to their origin and histopathologic features, retrorectal cysts are classified as squamous-lined (dermoid or epidermoid) cysts, postanal gut (tailgut) cysts, and rectal duplications (enteric or enterogenous cysts, enterocystomas). Described in this case report is an extremely unusual patient, a woman who simultaneously had a retrorectal cyst and an ovarian serous cystadenoma in addition to a long history of misdiagnosis and multiple unsuccessful surgeries.

Case Presentation: The patient was a 45-year-old female who presented with back pain, rectal fullness, constipation, and urinary symptoms. Upon her first pregnancy, a cystic pelvic mass had been misdiagnosed as an ovarian cyst. During the following 17 years, she had undergone several ineffective operations. The last CT scan and MRI studies revealed two separate noncalcified, unilocular, cystic lesions with well-defined borders in the retrorectal and retroperitoneal spaces. Two cysts were excised completely by a combined abdominoperineal approach. Pathological assessment revealed a dermoid cyst and an ovarian serous cystadenoma. No complications occurred during the 18 months of follow-up.

Conclusions: Coexistence of a retrorectal cyst and a serous cystadenoma is very unusual. Retrorectal cysts are rare entities that remain a difficult diagnostic and therapeutic challenge. Misdiagnosis and multiple unsuccessful surgeries are common. Complete surgical removal is the treatment of choice and requires a multidisciplinary approach in complicated cases.

Keywords: Rectum, Ovary, Cyst, Tumor, Epidermoid, Serous Cystadenoma
centers and even by unprofessional hands. The cyst was
not resected completely, and it recurred after each of these
procedures. After these unsuccessful surgeries, no other
major procedure was done, and the patient was followed
with transabdominal ultrasonography approximately ev-
ey six months. When the patient's symptoms became un-
bearable, the cyst was drained through a minimal perineal
incision between the coccyx and anus. In the last year,
her symptoms did not improve after perineal drainages. It
seemed that the cyst was not completely evacuated. A CT
scan and MRI studies confirmed two large, separated, non-
calcified, unilocular cystic mass lesions with well-defined
borders in the retrorectal and retroperitoneal spaces. On
a Ti-weighted MRI, the lesions were low signal, while on
a T2-weighted MRI, the lesions were high signal, indicat-
ing fluid content. There were no areas of heterogeneity or
irregularity that enhanced with contrast. The retrorectal
mass was located in the pelvic cavity behind the rectum
and the vaginal canal, compressing and anteriorly displace-
ing the uterus (Figure 1).

We decided to resect the cysts completely, because the
case was so complicated after 19 surgeries (laparotomies
and perineal drainage). A team of two general surgeons
experienced in anorectal surgery, a urologist, a neurosur-
geon, and two anesthesiologists operated on the patient
for eight hours.

For complete excision, we used a combined ab-
dominoperineal approach. At first, JJ stents were inserted
through cystoscopy to find and protect the ureters dur-
ing dissection, on account of the massive adhesions and
anatomic distortion caused by previous operations. Then
the laparotomy was done through a midline incision,
revealing a large cystic lesion, adherent to the small bowel
and ureters. The mass was carefully dissected and iso-
lated. It seemed to have originated from the left ovary,
and it contained clear fluid. The cyst was resected by sal-
pingo-oophorectomy.

The second mass in the retrorectal space was com-
pletely apart from the first cyst. Although exceedingly dif-
ficult because of adhesions and fibrosis from the previ-
ous surgeries, the cyst was dissected with special atten-
tion to avoid injuring the rectum, ureters, or pelvic ner-
vous plexus. For complete dissection of this cyst, the op-
eration continued with a perineal approach. A longi-
tudinal incision was made between the anus and the coc-
cyeal bone, the subcutaneous planes were divided, and
the lumbosacral fascia was exposed. The anococcygeal liga-
ment was transected. The retrorectal space was exposed by
transsection of the fibers of the levator ani. After complete
dissection of the cyst, it was removed with a tract that had
been created between cyst and skin in the site of the last
perineal drainage. The pelvic floor was then reconstructed
by suturing the fibers of the levator ani.

Upon histopathological examination, the first speci-
men consisted of the ovary containing a creamy brown
unilocular 9 × 7.5 cm collapsed cyst with a smooth intern-
al surface. The cyst was reported as “ovarian serous cy-
sadenoma.” The second specimen was a unilocular 12 × 6.5
× 8 cm cyst filled with yellow fluid and sebaceous mate-
rial; the inner aspect of the cyst had a creamy smooth sur-
face. Microscopically, this cyst was lined by stratified squa-
mous epithelium. Also seen were scattered sweet gland-
like structures, areas of erosion, granulation tissue forma-
tion, chronic inflammation, and fibrosis. The diagnosis
was “dermoid cyst.”

Postoperative recovery was complicated by respiratory
failure, but eventually the patient improved and was dis-
charged in generally good condition on the twelfth post-
operative day. No complications occurred during the 18-
month follow-up.

3. Discussion

The case described in the present report had two dif-
ferent problems simultaneously: a retrorectal cyst and an
ovarian serous cystadenoma. The coexistence of a retrore-
ctal and serous cystadenoma had been mostly unreported
in previous articles. In this case, the ovarian cyst may have
been a recently created mass that had been added to an
old problem, the retrorectal cyst. This was probably the
reason for the patient’s persistent symptoms despite four
drainages during the preceding year.

Unlike retrorectal cysts, serous cystadenomas are very
common benign tumors of the ovary that may occur in
any age group (8). Tumors of the ovary are common forms
of both benign and malignant neoplasia in women, but
about 80% are benign, and these occur mostly in young
women between the ages of 20 and 45 years. The benign
forms may be entirely asymptomatic, and there are occa-
sionally unexpected findings upon abdominal or pelvic ex-
amination or during surgery (9). Treatment of serous cy-
sadenomas can involve a cystectomy or oophorectomy, de-
pending on the amount of ovary involved (8).

Retrorectal dermoid cysts are rare developmental cy-
sic lesions that are thought to arise from caudal embry-
onic vestiges (4, 10). These benign lesions are lined with
stratified squamous epithelium and are filled with dense
muddy or fatty material. They can be differentiated from
epidermoid cysts with both gross and microscopic anal-
ysis, because they contain skin appendages such as hair
follicles, sweat glands, and tooth buds (4). Because these
cysts are sometimes multiple (4), we had initially misin-
interpreted the MRI findings of our patient as multiple retrore-

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tal cysts; but during the operation, we found two different origins for the cysts.

The symptoms of congenital retrorectal masses are often subtle and nonspecific (10). Many of these masses are incidentally discovered during routine gynecologic examinations (1, 6), as we saw for the first time in our case; however, some patients have symptoms secondary to infection in the cyst. They present with a history of recurring retrorectal abscesses and repeated operations for anal fistulae (6). A small group of patients having large cysts experience symptoms resulting directly from the compression of the surrounding structures where the mass causes pain, constipation, narrowed stools, or pollakiuria (4, 10).

Because retrorectal cysts are rare entities and have nonspecific clinical presentations, a large proportion of cases are initially misdiagnosed. These patients may present with a history of multiple unsuccessful drainage procedures (7, 11). It often happens that patients have been treated with an ineffective method for many months before they finally receive professional help (5).

Singer et al. reported seven patients with retrorectal cysts who had been misdiagnosed before referral. These patients had been treated for fistulae in ano, pilonidal cysts, perianal abscesses, psychogenic, lower back, post-
traumatic, or postpartum pain, and proctalgia fugax before the correct diagnosis was made. The patients underwent an average of 4.1 operative procedures (7, 12). Our case had several times been diagnosed and treated for a large ovarian cyst and even after correct diagnosis underwent multiple unsuccessful procedures. In addition to the rarity of the occurrence of retrorectal cysts, the most probable reason for such a situation is physicians’ lack of experience in their diagnostics (5). A history of multiple procedures should alert the clinician to the diagnosis of a retrorectal cyst. Once suspected, the correct diagnosis can be made with a physical examination and a CT scan or MRI before a definitive surgical procedure (7). Pelvic CT and especially MRI are the most important diagnostic tools when dealing with retrorectal cysts. Endorectal ultrasound can also be useful. It confirms the diagnosis and allows the precise determination of the size of the cyst and its relation to the rectum wall (5). All of these diagnostic tools provide crucial information for preoperative planning (2, 10).

The cornerstone of the treatment is complete surgical excision (6, 10). Chronic infection is the most frequent complication, occurring in 30%–50% of developmental cysts (3, 4). Because of the high predilection to infection, the preferred treatment for retrorectal cysts is complete excision. Removal is also recommended because of malignant degeneration arising in developmental cysts (6, 13, 14). Dermoidal cysts turn malignant in 10%–15% of cases (5). The approach to retrorectal masses can be abdominal, perineal, or a combination of the two, depending on the location and size of the lesion and its relationship with adjacent structures. If the mass does not extend above the level of the fourth sacral element, the perineal approach is the appropriate method. The abdominal approach is to be reserved for lesions whose lowest extent is above the level of the fourth sacral element, because this approach allows the best exposure of the pelvic structures (1, 15, 16). The combined approach is to be used for very large masses extending both proximally and distally to the fourth sacral element or for frankly malignant lesions with an infiltrative pattern that makes the isolation of the mass impossible by the perineal approach alone (10). We used the combined approach, because the retrorectal mass was extended below the level of the fourth sacral vertebral corpus in the MR images, and the transabdominal approach alone did not provide enough access to the two cysts.

Complete resection with negative margins is the standard surgical approach for retrorectal tumors. So in complicated cases, a multidisciplinary approach involving colorectal surgeons, neurosurgeons, possibly orthopedists, and radiation oncologists (for malignant tumors) is overemphasized in the literature (1, 17, 18). In spite of the benign course of the disease in the present case, we used a team of experts, because we expected probable adhesions, fibrosis, and anatomic distortion after several previous surgeries. The long-term outcome after resection of a retrorectal lesion depends upon the type of tumor and on adequate resection at the initial operation. For benign lesions, after complete surgical removal, the prognosis is good (10, 17).

3.1. Conclusions

The coexistence of a retrorectal cyst and a serous cystadenoma is very unusual. Retrorectal cysts are rare entities that remain a difficult diagnostic and therapeutic challenge. Misdiagnosis and multiple unsuccessful surgeries are common. Complete surgical removal is the treatment of choice and requires a multidisciplinary approach in complicated cases.

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