Presacral Cysts and Tumors

Douglas Wong, Josephine Tsai
Memorial Sloan Kettering Cancer Centre, USA

Retrorectal Tumors

The retrorectal space is a common site for embryologic remnants from which cysts and neoplasms may arise. These tumors are rare, but present interesting and challenging management decisions.

Anatomy

The retrorectal space is bounded superiorly by the peritoneal reflection; inferiorly by the presacral fascia or Waldeyer’s fascia; laterally by the lateral ligaments, ureter and iliac vessels; anteriorly by the fascia propria of the rectum; and posteriorly by the presacral fascia. Below the retrorectal space is the horseshoe-shaped suprarectal space.

Classification

Retrorectal masses can be divided into the following categories: congenital, inflammatory, neurogenic, osseous and miscellaneous. The following table lists the various tumors under each category.

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Osteogenic Sarcoma

Bone cyst

Ewing’s sarcoma

Chondroma

The most common neoplasm in the retrorectal space is the presacral teratoma. These tumors are derived from tissue of all three germ layers. The possibility of malignant degeneration is greatest during the growth period or during the early years. Mahour has reported that the frequency of malignancy in sacrococcygeal teratomas ranges from 10% to 50%. Of those neoplasms present at birth, if not treated, 7% will go on to become malignant by the fourth month. Malignant transformation in adult teratomas is about 30%. Teratomas rarely are malignant beyond age 20. Teratomas are more common in females, and are associated with anomalies of the vertebrae, urinary tract or anorectum.

Teratoma and Teratocarcinoma

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These lesions may be cystic or solid, and may contain a variety of tissue, including respiratory, nervous and gastrointestinal. The blood supply comes mainly from the mid sacral vessels. Retrectal teratomas may extend into the abdomen, downward into the perineum or posterior into the buttocks region.

These tumors may grow to a large size and cause dystocia. After birth, rapid growth may lead to ulceration, bleeding or urinary obstruction.

In adults, presacral teratomas are exceedingly rare. Unlike teratomas in infants, which are externally visible in 90% of cases, sacrococcygeal teratomas in adults are mostly confined to the pelvis. They may become secondarily infected and present as a presumed fistula-in-ano before being discovered as a retrorectal tumor. Teratomas may rupture into the rectum.

Treatment is complete resection, either by posterior approach or combined abdominosacral approach. Many authors advocate routine removal of the coccyx, as it may contain the nidus of totipotential cells. Failure to resect the coccyx has been associated with a high rate of recurrence. Presently, chemoradiation is not useful for sacrococcygeal teratomas.

Chordoma

Chordoma is the most common malignant neoplasm in the retrorectal space. It arises from the remnants of the notochord. In adults, the only notochordal remnant is the nucleus pulposus of the intervertebral disk. Chordomas do not appear to arise from the intervertebral disks, but from the vertebral bodies. The retrorectal region is the site of chordomas in 50% of cases. In the other half of cases, chordomas present at the base of the skull.

These tumors are more common in men, and are uncommon in individuals younger than 40 years. Patients note rectal or perineal pain that is worsened by sitting. Advanced tumors may present with fecal and urinary incontinence. Chordomas have been associated with colorectal tumors.

Chordomas are slow-growing, lobulated lesions, which can invade bone and other neighboring structures. Physical examination reveals a smooth extrarectal mass with normal, mobile mucosa overlying it. Plain films can reveal expansion of bone, destruction, trabeculation and calcification. CT scan can confirm the diagnosis, and is invaluable in planning resection. Yonemoto et al recommended MRI as an important preoperative study to evaluate possible infiltration into gluteal muscle. If the gluteal muscles are involved, a wide resection can be planned in advance. Biopsy is best avoided, except in extenuating circumstances as discussed later. Endorectal ultrasound is of value in assessing the rectal wall, as well as in identifying pelvic floor muscle involvement.

Treatment of chordomas is complete excision to include any biopsy tracts, since implantation along these tracks have been reported. Radiotherapy may be useful for...
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**Osseous**

Osteogenic sarcoma

Bone cyst

Ewing's sarcoma

**Incidence**

Retrorectal tumors are rare. Cleveland Clinic reports 50 cases over a 55-year period. The Mayo Clinic estimates the incidence to be about 1 in 40,000 hospital admissions.

They report 120 cases, of which 66% were congenital; 12%, neurogenic; 11%, osseous; and 11%, miscellaneous. Stewart et al combined reports for a total of 301 retrorectal tumors, of which 63% were congenital; 8%, inflammatory; 10%, neurogenic; 7%, osseous; and 12%, miscellaneous. Memorial Sloan-Kettering Cancer Center reported on 39 malignant tumors, of which 38% were chordomas; 15%, neurogenic; and 23%, miscellaneous.

**Congenital Lesions**

Congenital lesions account for more than 50% of all presacral tumors. Two-thirds of congenital lesions are developmental cysts.

**Developmental Cysts**

These cysts are a result of defective closure of the endodermal tube, which results in inclusions of skin and accessory appendages.

The epidermoid cyst is lined by stratified squamous epithelium. No skin appendages are found. Dermoid cysts have, in addition to squamous epithelium, sweat glands, hair follicles and/or sebaceous glands. Both types of cysts are well-circumscribed, and contain thick green-yellow fluid. They can communicate with the skin and appear as a postanal dimple. There is a 30% rate of infection, presenting as either a presacral abscess or mistakenly diagnosed as fistula-in-ano.

**Enterogenous Cysts**

These cysts are believed to be derived from inclusion of the developing hindgut. They are lined by intestinal epithelium, and contain layers of smooth muscle, a myenteric plexus and serosa. They may also be lined by squamous or transitional epithelium, as the terminal hindgut gives rise to both rectal and urogenital structures.

These cysts also have a tendency to become infected. Malignant degeneration in a retrorectal duplication cyst has been reported.

**Tailgut Cysts**

Tailgut cysts are thought to arise from remnants of the embryonic tailgut, and differ from teratomas in that not all three germ layers are represented. They are distinguished from epidermoid and dermoid cysts in that they contain some glandular or transitional epithelium. They are usually circumscribed and lined by a variety of epithelial types, including columnar, transitional and squamous.

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incompletely resected lesions. There is no chemotherapy regimen for chordomas.

The fact that complete excisions rates of only 30% to 53% are reported may account for 5-year survival rates ranging from 43% to 75%.

AN TERIOR S ACRAL M E NING O C E L E

The meningocele contains cerebrospinal fluid. It is more common in women. Patients may present with low back pain, constipation, headaches or meningits.

The "scimitar" sacrum on plain film and CT scan is pathognomonic for meningocele. The scimitar sacrum is one that has a rounded, concave border without bone destruction. MRI may replace CT scan and myelography for diagnosis. Biopsy should not be performed, since this runs the risk of meningitis.

Treatment is resection via either an anterior, posterior or combined approach. The anterior approach allows easier closure of the neck of the sac, but nerve roots are difficult to protect. The posterior approach allows good identification of the nerve roots, but obliteration of the neck may be difficult.

NEU RO G EN IC T U MORS

These tumors comprise about 5% to 15% of retrorectal neoplasms. Benign lesions include neurilemmomas and ganglioneuromas. Neuroblastosmas, schwannomas and ganglioneuromatosas are malignant lesions. Motor and sensory dysfunction usually involve a single peripheral nerve. Involvement of the nerve root may result in paraplegia. Neurogenic lesions are slow growing, and may reach a large size before detection.

The most common neurogenic neoplasm is the ependymoma. If the lesion is well-circumscribed and completely excised, long-term survival is good. Radiation treatment may be palliative.

O SSEO US T U MORS

Primary bone lesions in the retrorectal region are extremely rare. They may arise from bone, cartilage or marrow. Skeletal pain points to diagnosis of these tumors, which, when discovered, are usually well advanced. Osteosarcomas have a poor prognosis, since they are usually inoperable by the time of detection.

Bone cysts and osteomas tend to recur usually secondary to incomplete resection.

M IS C E L L EA N O US

Soft tissue sarcomas are rare, and carry a poor prognosis. One must also keep in mind that lymphoma, myeloma and metastatic carcinoma can occur in the retrorectal space. Pelvic ectopic kidneys may also present in this region.

Clinical presentation of Retrorectal Tumors

Symptoms Malignant lesions are more likely to produce symptoms. Pain is usually poorly localized as back, rectal or perianal pain. If the sacral plexus is involved, pain may be referred in the legs or buttocks. Pain is usually associated with sitting.

Infection may present with recurrent episodes of perianal suppuratin. Obstruction of the pelvic outlet may result in constipation, incontinence from paradoxical diarrhea or interference of the sphincter nerve supply, and dystocia. Bladder dysfunction may also be a part of the symptomatology.

Examination

Examination begins with inspection of the perianal area to evaluate for postanal dimple. Laxity of the anal sphincters may point to involvement of the pelvic floor innervation.

Most retrorectal tumors are palpable. Higher lesions can be felt by noting a sudden anterior angulation of the sacral curve. The mass must be assessed for its relationship to the sacrum and coccyx. Location will determine operative approach.

Cystic neoplasms may be difficult to palpate, particularly if they are not tense. Pressure over an anterior meningocele can cause a rise in fontanelle pressure in infants. In adults, the Valsava maneuver can demonstrate spinal canal continuity with the meningocele.

Sigmoidoscopy should evaluate the overlying mucosa, particularly an edematous submucosa, which may herald an infection.

Diagnoses

Plain x-rays are useful for lesions that arise from or invade into the sacrum. Anterior meningoceles carry the pathognomonic scimitar sign caused by a unilateral sacral defect and no bony destruction. Teratomas may present as bone or teeth in the pelvis. Chordomas can cause bony destruction.

A fistulogram of a chronic fistula may identify a retrorectal mass.

CT scans are the most important diagnostic tool in evaluating retrorectal tumors and planning operative strategy. MRI and myelography are helpful in delineating central nervous system involvement and in defining extent. Endorectal ultrasound is of value in assessing for rectal wall involvement, and in determining pelvic floor muscle invasion.

Angiography rarely changes the operative approach. It may be helpful, though, in deciding for preoperative embolization for very vascular tumors.

There is no role for preoperative biopsy of a retrorectal tumor that is deemed operable. Biopsy of solid malignant lesions will lead to tumor seeding along the tract; of cystic lesions, infection; and of meningocele, meningitis. Preoperative biopsy is warranted in inoperable cases so that adjuvant therapy can be planned. Biopsy is also indicated in cases in which the anticipated extent of surgery would be so great that it would be debilitating, and therefore considered only if the tumor was confirmed to be malignant. A parasacral or perineal approach is preferable to a transrectal approach, as a potential future resection can incorporate a parasacral or perineal biopsy site.

O PERAT ION

Distal retrorectal tumors such as presacral cysts can be managed surgically by the experienced colorectal surgeon. However, more extensive lesions are best managed by a multidisciplinary team comprised of a colorectal surgeon, and either an orthopedic surgeon and/or neurosurgeon.

The posterior approach is usually feasible if the examiner can reach the proximal extent of the tumor with his examining finger. The patient is placed in the prone jackknife position. A midline incision is made from above the posterior anal margin cephalad to the sacrum. The incision is deepened to the sacrum, coccyx, and anococcygeal ligament. The ligament is detached from the coccyx in addition to disarticulation of the coccyx from S5. Many authors advocate routine coccyx removal, secondary to improved exposure and lower rate of recurrence. All cystic lesions are believed to originate in the coccyx, so the coccyx should be removed en bloc with cystic lesions.

After incising the anococcygeal ligament, the levator ani is divided and the suprarectal space entered. At this point, the decision to remove sacral segments depends on the size and location of the lesion and the exposure needed. If sacral segments need to be removed, the gluteal insertions are divided on each side. The piriiformis muscles is divided. The sacrotuberous and sacrospinous ligaments are identified and cut. The lowest two sacral nerves may be divided without neurologic consequence. Some authors state that a unilateral S3 nerve division is also without neurologic deficit. However, bilateral S3 nerve division will result in incontinence. Removal of the lower portions of the sacrum will not result in sacral instability. Bleeding can be a problem until the sacrum is completely removed. One must be cautious with a high tumor, since the blood supply comes from above, and uncontrollable bleeding can occur in a deep, inaccessible hole. If, despite packing, hemorrhage is still ongoing, the patient will need to be repositioned and control obtained through the transabdominal approach.

After the tumor is removed, the wound is closed in multiple layers and drains left in place.

High lesions warrant a transabdominal and posterior approach. The abdominoperineal approach allows vascular control and good exposure. A midline abdominal incision is made. The left colon is mobilized from the retroperitoneum. The left ureter is identified and preserved. For cases in which anorectal preservation is planned, the posterior plane between the visceral and parietal fascia is sharply dissected to separate the rectum and its mesorectum from the presacral mass. This generally necessitates taking down the lateral stalks in order to mobilize the rectum posteriorly down to the level of the rectosacral fascia.

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**NEUROGENIC TUMORS**

These tumors comprise about 5% to 15% of retrorectal neoplasms. Benign lesions include neurilemomas and ganglioneuromas. Neuroblastomas, schwannomas and neurofibromas are malignant. Motor and sensory dysfunction usually involve a single peripheral nerve. Involvement of the nerve root may result in neurogenic lesions and metastatic carcinoma can occur in the retrorectal space. Pelvic ectopic kidneys may also present in this region.

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Update on Adhesion Prevention

Eric G Weiss
Associate Residency Program Director, Director of Surgical Endoscopy, Department of Colorectal Surgery, Cleveland Clinic Florida, USA

Introduction
Adhesions remain a significant source of morbidity and their prevention would significantly aid medical care. All abdominal surgical procedures have the potential for creating adhesions. In the absence of surgery, abdominal and pelvic infections and therapy, such as peritoneal dialysis, may incite the inflammatory cascade. Clearly, the optimal solution is that of prevention. Diminishing the deposition of fibrin and enhancing fibrinolysis without interfering with wound healing are the goals. This may be achieved primarily by four means: 1) mechanical bowel fixation (e.g. long tubes, suture pexy) to promote "friendly" or "benign" adhesions which will not lead to obstruction 2) systemic pharmacologic therapy (e.g. anti-inflammatory medications) 3) intraperitoneal therapy or barriers (e.g. carboxymethylcellulose, sodium hyaluronate, irrgiants) 4) local factors (e.g. surgical technique, foreign bodies), (Table 1). While none of these will completely prevent adhesions, several have been found to be promising in retrospective and prospective studies.

Table 1. Prevention Methods
Operative Techniques (local factors)
- Gentle handling of tissue
- Avoidance of ischemia
- Avoidance of infection
- Hemostasis
- Avoidance of foreign bodies

Mechanical Bowel Fixation
- Suture pexy (Noble or Childs-Phillips plication)
- Lumenal plication (Long tubes – ie.Baker or Gowen tubes)

Mechanical Barriers
- ePTFE (Gor-texTM)
- oxidized regenerative cellulose (Intracead R)
- sodium hyaluronate and carboxymethylcellulose (SeprafilmTM)

Pharmacologic (systemic)
- Steroids
- Nonsteroidal anti-inflammatory drugs

Mechanical bowel fixation techniques have been attempted internally and externally. The most common external techniques are tacking the bowel to the peritoneum (especially with stomas), and suture pexy of the small bowel loops and mesentery in an anatomically favorable position (Noble or Childs-Phillips procedure). While these have had some limited success in reducing the incidence of recurrent obstruction, they are associated with serious and frequent complications. These include enteric leaks, fistulas, sepsis and increasing the difficulty of reoperation. 1-4

The use of a long tube (Baker tube) to internally stent the bowel has also been evaluated. Again, the improvement is minimal, and complications include pneumonia, intussusception, and inability to remove the tube may occur. Close and Christensen have compared the Childs-Phillips plication to Baker tube placement, and to adhesiolysis alone. Overall, the incidence of recurrence for SBO was low for all three groups, although highest for the adhesiolysis group (6.5%). 5-6 The benefit of pexing or stenting is minimal so because of the potential associated complications, they are generally not recommended for uncomplicated adhesive bowel obstructions. Some cautious consideration may be given for patients suffering from multiple episodes. 1-4

Systemic Pharmacologic Therapy
Systemic therapy in an attempt to modify the inflammatory response has been investigated. Steroids, non-steroidal (NSAIDs) and aspirin have reduced the incidence of adhesions in vitro and in vivo. A recent prospective trial of an antiadhesional adjunct (Seprafilm®) also evaluated the use of steroids. There was a significant reduction in adhesions in patients receiving steroids. 7 Muzi et al compared the use of low dose and high dose aspirin to a control group. 8 The reduction in adhesion formation was greatest in the low dose aspirin group (46% compared to 77 high dose, 100% control). The peritoneal levels of thromboxane were reduced most in the low dose aspirin group, and prostacyclin was reduced only in the high dose group. These findings may help explain the superiority of the low dose aspirin. 9 Similar effects have been noted with steroids and NSAIDs. NSAIDs may have a significant role in the prevention of adhesions, in conjunction with mechanical bowel fixations.