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REVIEW

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The management of presacral masses in the adult

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Abstract Presacral (retrorectal) tumors are particularly rare in the adult. There is difficulty in the performance of diagnostic biopsy, and specialized imaging is required to plan surgical extirpation. This review assesses their incidence and classification as well as the principles involved in their diagnosis and surgical management.

Key words Presacral mass • Surgical management

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Introduction

Presacral (retrorectal) tumors in the adult are rare; in general, the average coloproctologist has limited experience of their treatment. There is confusion about their origin and histological classification and although the majority are benign, they present a number of problems in diagnosis and management. Clinical diagnosis is often delayed because of their vague symptomatology and there is debate and difficulty concerning preoperative biopsy of these lesions. Definitive surgical excision may be hampered by relative inaccessibility, tumor size and fixity, intraoperative blood loss, or the experience of the surgeon with certain specialized approaches [1].

The retrorectal space is a potential space limited anteriorly by the fascia propria of the rectum and posteriorly by Waldeyer's rectosacral fascia [2]. Laterally it is limited by the iliac vessels and the ureters and extends along the lateral ligaments of the rectum, communicating superiorly at the level of the peritoneal reflection (S2–3) with the retroperitoneal space. Waldeyers's fascia effectively isolates the presacral space from the supralevator region, forming a horse-shoe-shaped region closed anteriorly by Denonvillier's fascia [3, 4]. It normally contains loose connective areolar tissue, the middle sacral, iliolumbar and middle hemorrhoidal vessels, assorted lymphatics, branches of the sacral plexus and occasionally, isolated embryological cell rests. The area represents a region where a number of embryological closures occur during development, forming the potential for neoplasms incorporating notochordal, cloacal, neurogenic and other developmental anlages [5, 6].

Epidemiology and classification

The first reported case of a presacral teratoma in an adult was by Emmerich in 1847 [7]. These tumors are particularly rare in the adult population, occurring in 1 of 40 000 hospital admissions, although they represent one of the commonest forms of

malignancy in the infant. Adult tumors differ from those of the infant in that the latter frequently present as externally visible sacrococcygeal masses which show malignant transformation over months if left untreated [8, 9]. The majority of presacral masses in the adult are benign, have a female predilection and are asymptomatic, presenting to the coloproctologist after being found incidentally on rectal examination. They are found in less than 0.02% of proctosigmoidoscopies performed in specialized clinics [10]. It has been suggested that malignancy is more common in men, although the literature is somewhat difficult to interpret since infantile and adult tumors are often categorized together as is the inclusion of malignant sacral and neurological tumors which secondarily involve the presacral space; most notably the malignant chordoma [11, 12].

The classification of these adult tumors is also confused since the embryology of the region is poorly understood. Traditionally, their classification has been separated into congenital, inflammatory, neurogenic, osseous, specific soft-tissue masses and miscellaneous tumors, such as myeloma, metastatic deposits and arteriovenous malformations [13, 14]. It is more practical to exclude inflammatory disorders in the presacral space and to combine miscellaneous and mesenchymal soft-tissue masses as we have done (Table 1), dividing neoplastic lesions into cystic and solid where the commonest cystic lesions represent developmental cysts; most notably dermoid and epidermoid cysts, tailgut cysts, cystic hamartomas, cystic teratomas, neurenteric cysts and enteric duplications [15]. Solid lesions include chordomas, primary mesenchymal and neuro(endocrine) tumors and metastatic deposits. These classification systems remain confusing because of the frequent reference in adult categorization of simple dermoid and epidermoid tumors as infantile-type sacrococcygeal tumors despite the fact that for a pathological diagnosis of the latter condition, remnants or origin from all primitive germ cell layers must be identified [16]. The inclusion of tailgut cysts specifically refers to benign developmental remnants and excludes duplication cysts, those with an intestinal epithelial lining, tumors with a muscular or serous coat or masses presenting with single simple linings, such as ectodermal squamous components which define dermoids, epidermoids and dermoid-related adnexal structures [17].

We believe that stricter definitions will effectively exclude confusing terms which appear in the literature such as postanal gut cysts and retrorectal cystic hamartomas, both of which are ambiguous terms [18]. Tailgut cysts have traditionally been associated with sacrococcygeal anomalies as classically described by Currarino et al. [19] in their triad of presacral cysts, sacrococcygeal anomalies and anorectal malformations. A review of the literature by Head et al. [20] in 1975 revealed only 71 cases of presacral tumors in adult patients; a recent update by Bull et al. [21] in 1999 added an extra 15 cases. Given these stricter criteria for the definition of adult presacral tumors, we have identified an additional 131 cases from the literature, including 30 sacrococcygeal tumors, 29 tailgut cysts, 18 benign and malignant neural and neurilemmal tumors, 2 arteriovenous malformations, 23 dermoid/epidermoid cysts, 15 myelolipomas, 4 carcinoid deposits and 10 miscellaneous masses (including 5 mesenchymomas, one granular cell tumor and 4 metastases; 2 from colonic adenocarcinoma and 2 hematopoietic in origin). One other paper reported 28 cases of adult presacral tumor, although details regarding the patients are not available from this source [22].

Table 1 Classification of presacral (retrorectal) neoplasms

Etiology	Tumor type
Congenital or developmental	Developmental cyst Epidermoid cyst Dermoid cyst Tailgut cyst Teratoma Teratocarcinoma Enteric duplication
Neurogenic	Neurofibroma Neurofibrosarcoma Neurilemmoma Ependymoma Neuroblastoma Chordoma Anterior sacral meningocele
Osseous	Osteoma Osteochondroma Osteogenic sarcoma Simple bone cyst Giant cell tumor Ewing's sarcoma Chondromyxosarcoma Aneurysmal bone cyst
Miscellaneous	Metastatic carcinoma (Myelo)lipoma Liposarcoma Fibroma Fibrosarcoma Hemangioma Pericytoma Lymphangioma Extra-abdominal desmoid tumor Carcinoid tumor

Clinical presentation and investigation of presacral masses in the adult patient

The possibility of a presacral mass should be considered in patients presenting with a recurring retrorectal abscess, repeated anal fistulas (or an inability in such patients to delineate typical cryptoglandular sepsis), the presence of postanal dimpling and evidence of fixation or fullness in the precoccygeal and/or presacral region on clinical examination. Although at least half of the presacral tumors presenting in adults are asymptomatic and the majority are benign, it is generally recommended that these lesions be removed because of their propensity to become malignant over time [23]. Given the natural history of the anterior sacral meningocele to become infected, it is also recommended that these lesions be electively resected [24, 25]. The difficulty of preoperative representative biopsy, the tendency for cystic lesions to become infected (particularly after biopsy),

difficulties in operative access by different approaches to the limits of these tumors, pelvic dystocia with large presacral masses in the parturient female and their frequent recurrence after limited excision or incision imply that an image-directed approach is required in their surgery. Poorly planned, repeated operations for recurrent masses in this region carry a considerable risk of fecal incontinence in some patients.

Presacral tumors are typically slow growing, producing symptoms by pressure and/or displacement of pelvic structures in the narrow pelvic cavity. Symptoms and signs suggestive of nerve root pain, large tumor size, urinary or bowel complaints and even apparent bony fixation do not necessarily signify malignancy, although urinary and fecal incontinence are more indicative of malignant transformation. The single most reliable indicator for malignant transformation is sacral destruction [26].

Imaging recommendations for presacral masses

Plain X-rays in these patients are frequently unhelpful but in some cases, they may show soft tissue masses, calcification

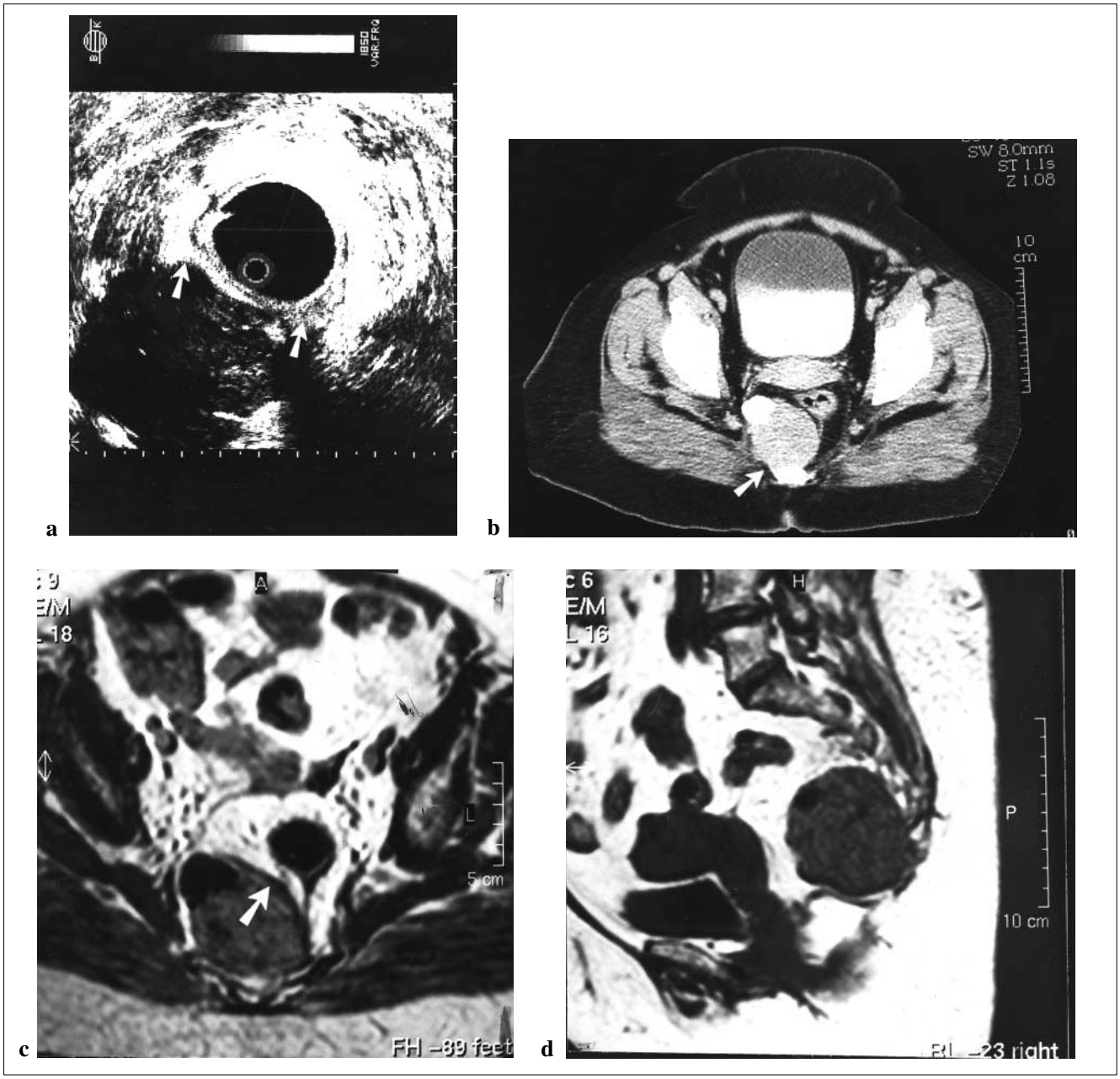

Fig. 1 a Endorectal ultrasound (ERUS) of the presacral mass, showing infiltration of the echogenic rectal muscularis (*arrows*). **b** CT scan showing axial views of the mass (*arrow*). Information with this modality is limited in discerning the relationship between the tumor and the rectum or the exact tumor level. **c** Axial MRI showing a clear edge between the heterogeneous mass and the rectal wall (*arrow*). **d** Sagittal MRI showing the level of the tumor extending to the tip of the coccyx with a clear boundary between the posterior aspect of the mass and the presacral fascia

and displacement or destruction of the sacrum or coccyx. Plain radiology frequently underestimates the extent of sacral damage since the sacrococcygeal region is difficult to examine due to overlying bowel gas and sacral foreshortening as a result of its normal curvature. Lateral views often show poor detail because of large volumes of soft tissue; although indirect information may be obtained from a barium enema or an intravenous urogram, however these are relatively crude methods of assessment [27, 28]. These facts imply an important role for both computed tomography (CT) and magnetic resonance imaging (MRI). CT is useful in delineating the relationship of the presacral mass to important structures such as ureters, uterus, bladder and rectum; this is vital in surgical and radiotherapeutic planning and in excluding the diagnosis of anterior sacral meningocele, for which preoperative biopsy is contraindicated [29]. MRI is highly recommended in presacral tumors, because of its multiplanar capacity and improved soft-tissue resolution, defining a plane between the tumor and the anterior presacral fascia in sagittal views. The latter is vital in decision-making regarding the surgical approach and the role of limited sacrectomy [30, 31]. In those cases where sacral invasion is identified, MRI defines its extent and the potential resectability of the tumor, where proximal extension into the S1–2 disk space defines inoperability. MRI is also more sensitive than CT at spinal imaging, showing associated cord anomalies such as variant spinal dysraphism and sacral arachnoid cysts [32]. More recently, endorectal ultrasound (ERUS) has been employed in solid or partially solid retrorectal masses where preoperative image-guided biopsy is contemplated. Figure 1 shows the CT, MRI and ERUS appearances of an adult presacral dermoid cyst that was completely excised by a combined abdominotrans-sacral approach in our unit. In this case, no further information was provided by this form of imaging although it showed the relationship of the mass to the muscularis propria of the rectum [33].

In general, we favor the preoperative use of ERUS and MRI, in particular since they both provide information regarding peritumoral planes which will govern the most desired surgical approach.

Preoperative biopsy of presacral masses

The role of preoperative tumor biopsy is controversial. Although much of the information regarding CT-guided approaches to the space has been provided by experience with sepsis following colorectal surgery in this region [34, 35] or in the diagnosis of presacral tumor recurrence after abdominoperineal excision and radiotherapy [36], access to this region is difficult. Moreover, there is a reported incidence of serious and even fatal infections, particularly in predominantly cystic lesions. The routes of access for biopsy are limited because of the sacrum itself. These approaches include a transgluteal approach via the greater sciatic foramen, a transrectal approach using endorectal US and dorsal transacral, parasacral and precoccygeal approaches [37–40]. Risks from biopsy include bleeding from damage to the middle sacral artery, injury to the anococcygeal ligament or the sympathetic ganglion impar and post-procedural insertion site pain or neuralgia. The advantage of CT guidance is the use of stacked multiplanar reconstructions from images acquired by helical CT of the pelvis. Although sagittal reconstruction of the plane of the traversing needle permits accurate measurement of the distance from the tip of the coccyx to the caudal aspect of the lesion by computerized vector plotting, in some cases lateral and presacral approaches can involve a prohibitive distance of travel through buttock fat in obese patients. We only recommend preoperative biopsy of solid lesions in certain circumstances, namely for patients in whom there is sacral invasion suggestive of malignancy (where there is the potential for postoperative radiotherapy), and in clearly malignant cases presenting as locally advanced disease, where non-operative therapy only is to be employed [41–44].

Surgical approaches and recommendations

The presacral tumor presents several operative difficulties because of limited access to the caudal component of the tumor via the abdominopelvic approach and because of poor vascular control when the approach is posterior. Decisions regarding the operative approach must be based on the size of the tumor, its rostral and caudal extent, involvement of contiguous vital structures, whether the case is a primary extirpation or a recurrence and on the familiarity (and comfort) of the surgeon with either pelvic or post-sacral anatomy. Preoperative bowel preparation is mandatory and it is advisable to discuss the case with the pathologist before surgery in order to arrange frozen section facilities. In certain circumstances where there is significant sacral invasion or preoperative neurologic symptomatology, a combined approach between the coloproctologist and a neurosurgeon is recommended.

Alternative operative approaches include posterior (trans-sacral), trans-sphincteric, transrectal, transvaginal and combined synchronous or staged abdominotrans-sacral approaches.

Abdominopelvic vs. post-sacral approaches

Anterior sacral meningoceles are usually more readily approached from an abdominopelvic perspective, however, operative decision-making is dependent more upon the

locale and size of the tumor than on its consistency. Abdominal approaches are recommended for high tumors commencing at or above the sacral promontory, providing direct visualization of the middle sacral artery, the presacral veins and the presacral nerves [45]. Wholly posterior approaches (the Kraske operation) are reserved for low lying tumors and infected cystic lesions whose rostral extent begins at the level of the mid-sacrum and extends as far as the tip of the coccyx or ano-coccygeal raphe, or where there is intimate involvement of the pre-coccygeal space. This posterior approach to the low-lying pelvic presacral mass has been modified for this lesion following success with the resection of certain types of mid-rectal cancers and large sessile villous adenomas [46–48]. Here, identification of the sacrum, coccyx, and the ano-coccygeal ligament is straightforward using a midline, curvilinear or horizontal incision. The coccyx may be disarticulated if necessary, with or without gluteus maximus detachment and attendant distal sacrectomy where appropriate, i.e. below the S4 level. However, extensive isolated resection of large tumors with this approach is not advised as there is no possibility of proximal vascular control and intraoperative hemorrhage can be catastrophic. The coccyx is resected in cystic lesions using this approach since the lesions may have an intimate bony connection and coccygectomy avoids tumor recurrence. Here, the incision should be made at the sacrococcygeal junction as close to the coccyx as possible in order to avoid injury to the lateral pelvic nerves. If the tumor is large, this procedure can be associated with an extraperitoneal iliac incision for vascular isolation. If the mass is closely adherent to the rectal muscularis, a digit placed intrarectally during coccygectomy and presacral dissection can be useful, as there is no peritoneum at this level.

Specialized alternative surgical approaches

A variation of the posterior approach uses a York-Mason style trans-sphincteric access where there is extension of the tumor into the rectal wall, although function following this procedure is often relatively poor [49, 50]. The combined abdominosacral approach which had previously been described for mid-rectal cancers was translated by Localio et al. [51, 52] into a synchronous procedure with preliminary vascular control and the potential for a diversionary colostomy; the procedure was originally developed because of the poor results obtained with either an isolated pelvic or posterior procedure for chordoma. The rectum is dissected from the tumor as far as the levator ani and, when possible, the most prominent sacral veins are ligated. The sacral incision is enlarged to join the abdominal incision with division of the sacrotuberous, sacrospinous and lower sacroiliac ligaments and detachment of the piriformis muscle and transection of the sacrum. Further advantages for these larger abdominosacral tumors include an ability to secure the ureters and rectum as well as the ability to ligate the mid-sacral artery under vision. Every attempt should be made to preserve at least one side of S2 to avoid urinary retention and a neurogenic bladder. We recommend this approach only when there is limited access to the caudal aspect of the tumor abdominally, when there is an extensive sacral involvement, and in recurrent cases [53].

The transrectal approach should only be considered in cases of ruptured transrectal cysts and rarely for a non-cystic dumb-bell lesion extending through the rectal muscularis propria which is sufficiently low enough for this approach [54].

Modifications of this approach for very low tumors include an intersphincteric procedure with entry into the retrorectal space via the intersphincteric plane, akin to the approach described by Parks for postanal repair [55], or a transvaginal incision (the Schuchardt-Schauta incision), when the tumor is sufficiently low and veers off the midline, lying between the rectal muscularis and the vaginal wall [56, 57]. These approaches avoid the possibility of sacral nerve injury and postoperative urinary retention as well as inadvertent rectal perforation, but they risk damage to the anal sphincters.

The views concerning surgical decision-making as presented in this paper have been developed in our unit as a result of our experience with these lesions. In summary, decisions regarding the surgical approach to presacral masses encountered in adult coloproctological practice are dependent on an accurate preoperative knowledge of the rostral and caudal extent of the lesion, its relationship to the sacral bony mass, its size and involvement of other pelvic structures (most notably the rectum, rectovaginal septum or vaginal wall), the presence of a predominantly cystic component or whether the surgery is for a recurrent mass. Complex embryological classifications appear to offer limited value in assisting the operative decisions regarding these rare lesions. Most often, presacral masses are discovered incidentally on clinical examination but they require specialized imaging (including sagittal MRI when possible) to define whether there is a plane of cleavage between the mass and the sacrum or in certain cases, the presence of sacral and coccygeal destruction and attendant variants of spinal dysraphism. ERUS offers limited value in defining these masses although it predicts the likelihood of rectal muscularis involvement, affecting the decision regarding operative approach. Preoperative biopsy is generally not recommended except in cases where non-operative management only is to be used. Abdominopelvic approaches are limited in their ability to get to the caudal extent of low-lying tumors but provide an opportunity (in association with staged or synchronous abdominotrans-sacral procedures) to carry out attendant rectal resection and to secure vascular control for larger lesions. Isolated post-sacral approaches should only be used for very low, smaller tumors and may be accompanied by a coccygectomy, which is recommended when the lesions are cystic in nature.

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