Presacral Tumors and Cysts in Adults*

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THE DISCOVERY of a presacral mass in an adult raises a number of questions for the examining physician: What are the possible causes of this mass? Is it malignant or benign? What diagnostic procedures are in order? What consultation should be obtained? What surgical approach, if any, should be considered? Our interest in this subject was stimulated by just such a discovery.

This paper presents the approximate total of presacral tumors and cysts in adults encountered over a 30-year-period in a major metropolitan area. Medical records from all of the area's major hospitals were examined. Follow-up data were obtained from the patients' physicians and by patient contact, where possible, or from medical records. In nearly all instances, tissue was obtained for examination.† Sixty-three cases were accepted for this study.

Anatomy and Pathogenesis

The presacral space, often referred to as the "retrorectal" space, is a "potential" space that lies anterior to the sacrum and coccyx and posterior to the rectum. The inferior boundary is the levators ani and coccygeal muscles. The superior boundary is the pelvic peritoneal reflection, and the lateral limits are defined by the ureters and iliac vessels (Fig. 1). The space contains branches of the sacral and sympathetic plexuses of nerves, as well as the middle sacral, ileolumbar and middle hemorrhoidal vessels and lymphatics.[9]

A study of the caudal end of the embryo is most instructive when considering the pathogenesis (*i.e., embryogenesis*) of the various types of tumors and cysts that can develop in the presacral space (Fig. 2). A number of developmental closures occur in this area, and many types of embryonic tissue are present.[6, 10, 14, 27] One can appreciate that the postanal gut could be pinched off, and leave a remnant that would form an endodermal cyst. Notochord tissue, as well as neurogenic tissue rests, can be anlage for developmental problems in the adult. A signal paper on this subject, published in 1938 by Gius and Stout,[6] concluded: "The presence of cysts in the perineal region can be explained on an embryologic basis," a statement with which we agree.

Present Series

We have followed the classification of these tumors and cysts as presented in 1949 by Lovelady and Dockerty[17]: Congenital, inflammatory, neurogenic, osseous, and miscellaneous. Table 1 lists the differential diagnosis of presacral lesions reported in medical literature, as well as the distribu-

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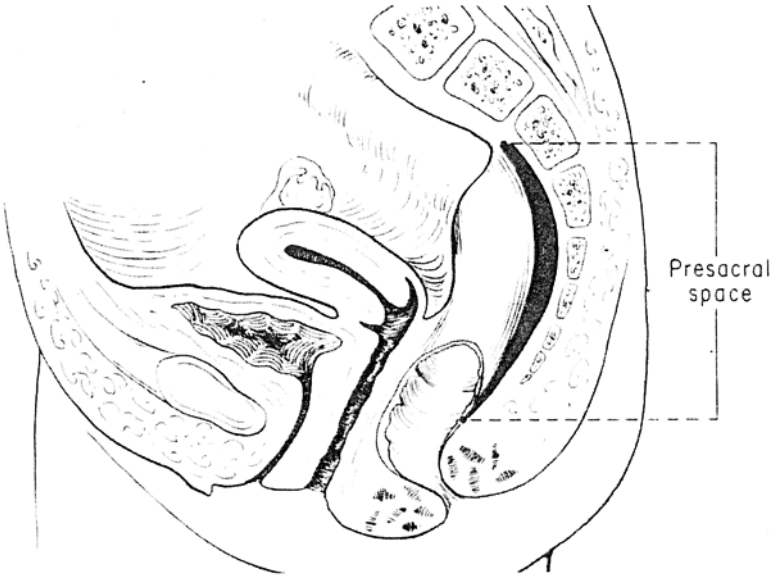

FIG. 1 *(above)*. Presacral space.

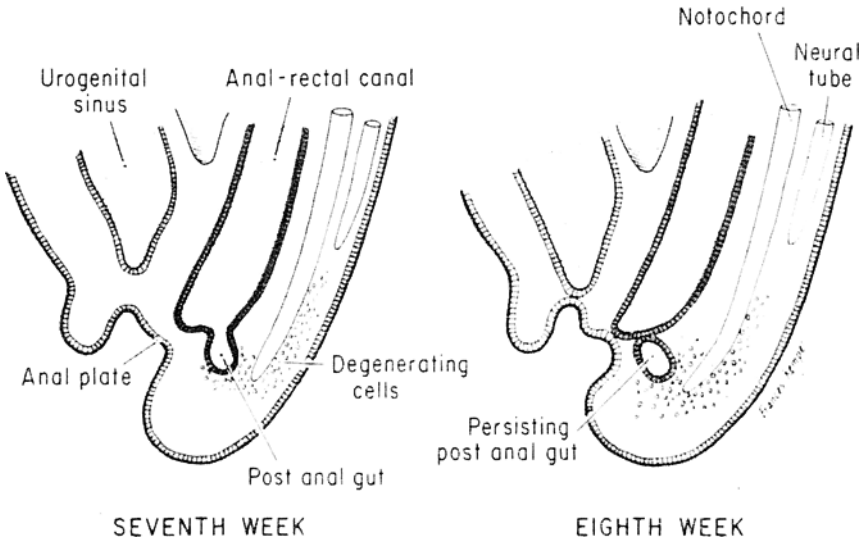

FIG. 2 *(below)*. Caudal end of embryo.

tion of types of lesions found in the present series. Thirty-seven of the 63 cases, or 58 per cent, were benign, and 26, or 42 per cent, were malignant. All patients were Caucasian except one American Indian woman who had a teratoma; 73 per cent were found in females and 27 per cent in males.

Congenital Lesions

Table 2 outlines the group of congenital lesions. Thirty-five cases in the congenital group accounted for 55 per cent of our series.

Developmental Cysts — Benign: There were 26 developmental cysts in the series: 16 mucus-secreting, no dermoids, one epi-

TABLE 1. *Differential Diagnosis of Presacral Lesions,[3, 9, 21] Including Present Series*

Differential Diagnosis	Present Series	
	Number of Patients	Per Cent
Congenital	35	55
Developmental cysts (epidermoid, dermoid and mucus-secreting cysts and teratoma)	26	
Chordoma	6	
Teratocarcinoma	2	
*Adrenal rest tumor	1	
Anterior sacral meningocele	—	
Duplication of rectum	—	
Inflammatory	3	5
Foreign-body granuloma	1	
Perineal abscess	2	
Internal fistula	—	
Pelvirectal abscess	—	
Chronic infectious granuloma	—	
Neurogenic	6	10
Neurofibroma and sarcoma	2	
Neurolemmoma	1	
Ependymoma	1	
Ganglioneuroma	2	
Neurofibrosarcoma	—	
Osseous	3	5
Osteoma	1	
Osteogenic sarcoma	1	
*Simple bone cyst, sacrum	1	
Ewing's tumor	—	
Chondromyxosarcoma	—	
Aneurysmal bone cyst	—	
Giant-cell tumor	—	
Miscellaneous	16	25
Metastatic carcinoma	9	
Liposarcoma	2	
Hemangioendothelial sarcoma	1	
*Lymphangioma	1	
*Extra-abdominal desmoid tumor	1	
Plasma-cell myeloma	1	
Malignant tumor ? type	1	
Lipoma	—	
Fibroma	—	
Fibrosarcoma	—	
Leiomyoma	—	
Leiomyosarcoma	—	
Hemangioma	—	
Pericytoma	—	
Endothelioma	—	
TOTAL CASES	63	

* Added by this series.

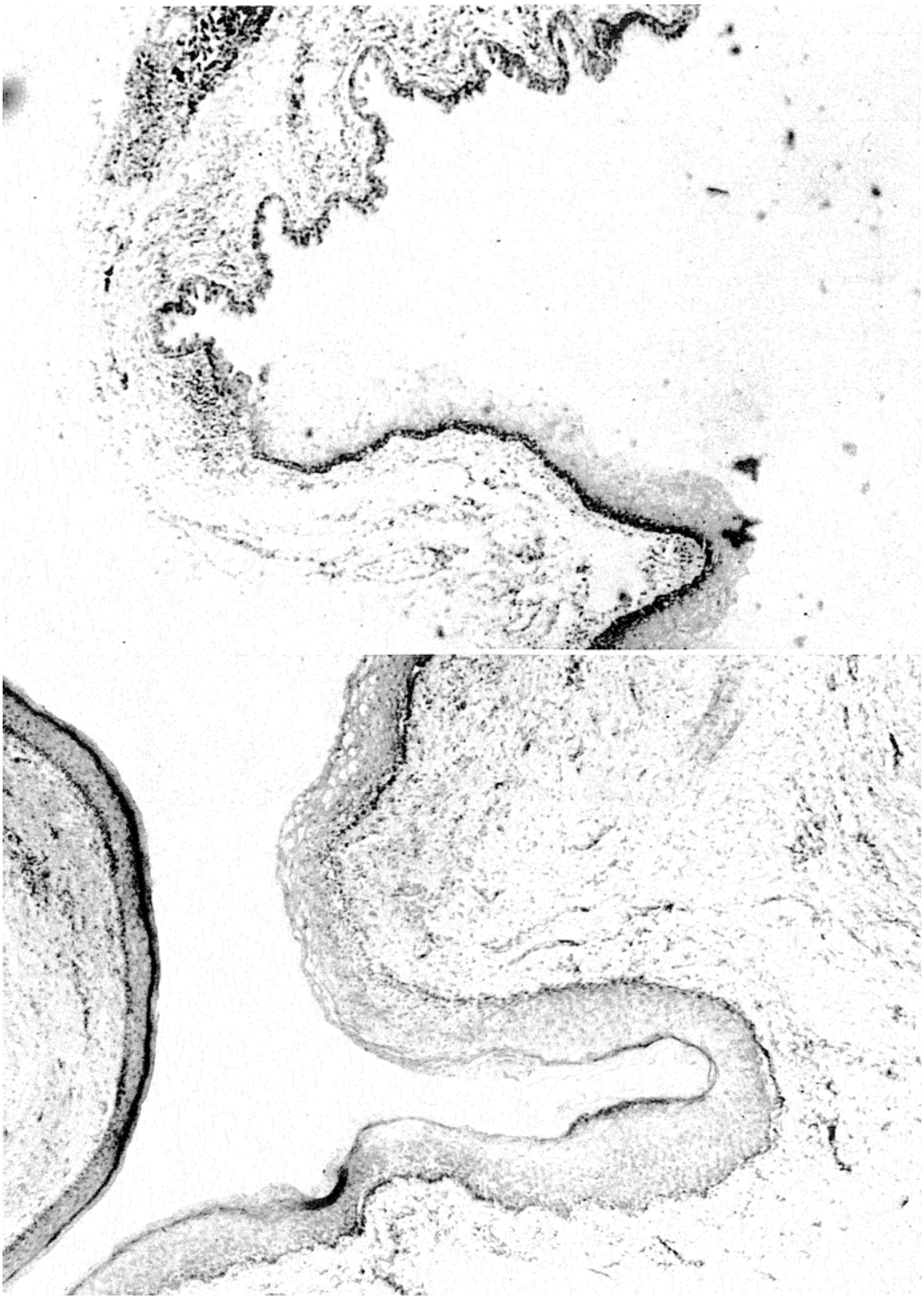

FIG. 3 *(above)*. Photomicrograph of mucus-secreting presacral cyst (×85, reduced from ×100).

FIG. 4 *(below)*. Epidermoid presacral cyst (×85, reduced from ×100).

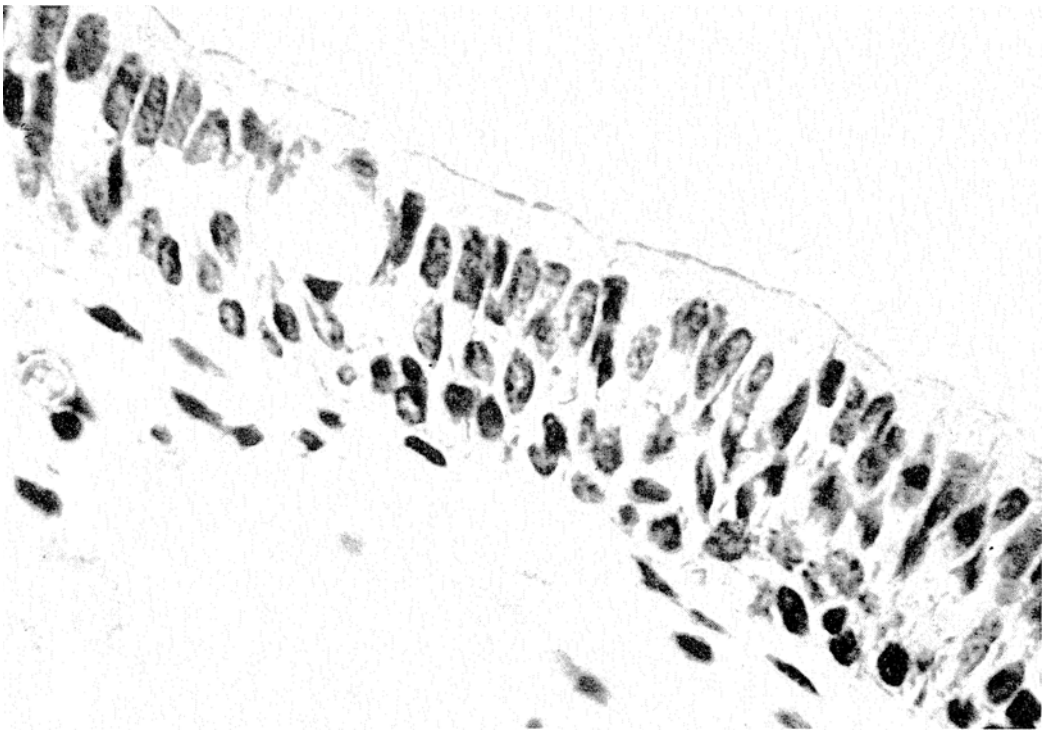

FIG. 5. Presacral teratoma—respiratory epithelium with cilia (×340, reduced from ×400).

dermoid, two teratomas and seven indeterminate cysts.

Confusion concerning the terminology of these congenital developmental cysts exists in the literature and in clinical practice.[4, 5, 7–9, 26] Many times we encountered the pathologic diagnosis of a "dermoid" when, indeed, only mucus-secreting epithelium was found. Classically, basic terminology defines a dermoid as having squamous epithelium (ectoderm) plus skin appendages (sweat glands, hair follicles, tooth buds, etc.). In none of the cases did we find one such skin appendage!

Mucus-secreting cysts are usually lined with columnar mucus-secreting epithelium; however, squamous or transitional epithelium may also be present (Fig. 3). They usually present as multiloculated cysts with one or two "parent cysts" and multiple "daughter cysts" present. They are thin-walled and contain a clear to light green mucoid material. Mucicarmine stains reveal a characteristic mucus-secreting epithelium. Edwards[2] makes the point that these lesions should be called "cyst hemartomas" to emphasize their congenital origin. We prefer, however, to use the term "mucus-secreting developmental cysts."[8, 26]

Epidermoid cysts have squamous epithelium present, but lack skin appendages. We found one such case (Fig. 4).

TABLE 2. *Congenital Lesions*

A.	Developmental cysts—benign		26
	1. Mucus-secreting cysts	16	
	2. Dermoid	—	
	3. Epidermoid	1	
	4. Teratoma	2	
	5. Indeterminant	7	
B.	Developmental cysts—malignant		2
	1. Teratocarcinoma	2	
C.	Chordomas		6
D.	Adrenal rest		1
	TOTAL		35

TABLE 3.

Presence of a presacral mass
Presence of a sinus or dimple
History of recurrent perianal abscess or sinus
History of many rectal operations
Hair or sebaceous material extruding from a sinus
Patient is female
Inability to find an anorectal origin for the symptoms

A teratoma is characterized as having two or three germ-cell layers present, and may contain hair, brain, cartilage, kidney, liver, etc. Two cases met the criteria to be classified as teratomas. Both cases demonstrated respiratory epithelium with characteristic cilia (Fig. 5). The associated mucous glands were reminiscent also of respiratory mucous glands. Other sections revealed multiple intertwined sections of smooth muscle characteristic of the gastrointestinal tract.

Middeldorpf,[20] in 1885 was the first to describe a mucus-containing cystic postanal teratoma; his patient was a 1-year-old child. That teratoma contained a fragment of intestine. Since then, many physicians have referred to all tumors in the presacral space as Middeldorpf tumors. According to strict usage, however, only teratomas in this region should be called Middeldorpf tumors.

There were seven indeterminate cysts in this series. These were probably mucus-secreting cysts that became so inflamed that a definitive microscopic identification could not be made. Most of these patients had had many rectal operations for an unrecognized cyst.

Clinical Data Regarding Presacral Cysts

A review of the clinical data regarding the presacral developmental cysts emphasized several important points (Table 3).[8]

The most important feature, in addition to a presacral mass, is the presence of a characteristic funnel-shaped postanal dimple. This dimple is found in the posterior midline of the anal canal, below the dentate line, and is easily confused with a fistulous opening (Fig. 6). It may or may not communicate with a presacral cyst. In our series, this dimple was recognized nine times out of 26 cases. It should be noted that these were patients of colorectal surgeons who were aware of and looking for the characteristic dimple.

The mean age at operation was 44 years. The mean duration of symptoms before definitive surgery was 3.7 years, a delay we attribute mainly to physician failure to diagnose. Twenty-two of the 26 patients were female, and four were male. A female preponderance has recently been emphasized.[7, 23] All but one of the mucus-secreting cysts were in female patients. The diameter of the cysts ranged from 2 to 15 cm. Six patients were asymptomatic, and the lesions were found on routine examination. A deformed coccyx was noted in six patients, and eight had undergone previous rectal surgery. Four patients had low back symptoms severe enough that orthopedic or neurosurgic consultation had been obtained. Three patients had another midline lesion (two vaginal cysts and one carcinoma of the thyroid). The initial impression that many of these cases would be associated with a high incidence of another midline lesion did not prove to be correct. None of the patients in this series were related, although a familial tendency of the epidermoid variety of cyst has been reported.[26]

Teratocarcinomas: The malignant potential of teratomas is stated to be 9 to 29 per cent.[10, 11] There were two teratomas and two teratocarcinomas in this series. One teratocarcinoma was in a 70-year-old woman who was found to have an asymptomatic presacral mass while upper abdominal surgery was being performed. This mass was removed through the abdomen and proved to be a teratocarcinoma with a one-sided development towards a thyroid follicular carcinoma (Fig. 7). This tumor

was incompletely removed by this approach and subsequently recurred, producing pelvic and sacral pain. The second surgical approach was transsacral, and again the mass was incompletely removed. The patient received cobalt therapy, but was subsequently lost to follow-up.

The second patient was a 49-year-old woman who developed pain when sitting. The physical examination revealed a large presacral mass. An abdominal approach was utilized to remove the tumor, which proved to be an embryonal adenocarcinoma developing from a respiratory epithelial component of the teratoma. A nine-year follow-up reveals no recurrence.

Chordomas: There were six chordomas in our series. The average age of this patient group was 50 years (range 39 to 70 years). There were five women and one man. This is at variance with the large referral hospital experience, where chordomas make up the largest number of the congenital lesions.[1, 3, 15, 18, 19, 25] Also, their patients were predominantly male.

Adrenal Rest Tumor: One previously unreported lesion is added to the differential diagnosis of lesions in this congenital category, an adrenal rest tumor (Fig. 8). The patient, a 64-year-old woman who had rectal discomfort, was found to have a mass in the presacral space. The lesion measured 8 x 8 x 8 cm and was completely removed through a transsacral approach. Two years later, there is no evidence of recurrence.

Inflammatory Lesions

Three inflammatory lesions, representing 5 per cent of our series, represented unusual diagnostic problems. The first patient had a presacral abscess that arose from occult perforated sigmoid diverticulitis. The second had a foreign-body granuloma subsequent to injection of phenol and mineral oil into her hemorrhoids years before (Fig. 9). She subsequently had a presacral mass that was found to be a lipogranuloma.

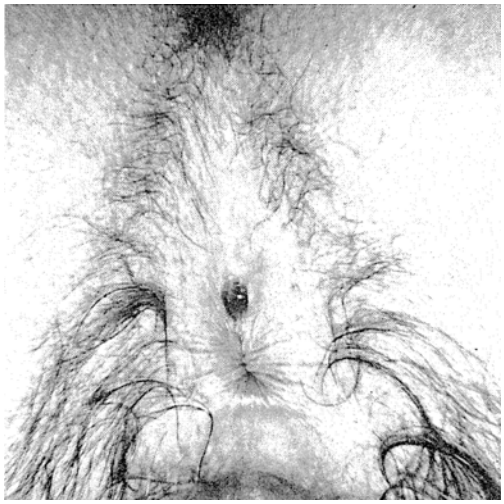

FIG. 6. Characteristic dimple associated with a presacral cyst. (Courtesy of Stanley Goldberg, M.D.)

The third patient had undergone a subtotal colectomy for ulcerative colitis years before, and subsequently had a presacral mass. This was found to be an abscess cavity filled with pus and granulation tissue, which was drained and subsequently healed.

Neurogenic Tumors

Six patients, averaging 33 years of age, had neurogenic lesions (Table 4). This group of patients seemed to have an unusually favorable prognosis, and except for the patient with the ependymoma, they are all alive and well with no evidence of recurrence. The patient with the ependymoma is a paraplegic, and for the past two years, his status has been stable.

Osseous Lesions

Three osseous lesions accounted for an incidence of 5 per cent of this series: one osteoma, one osteogenic sarcoma, and one simple bone cyst of the sacrum. The simple bone cyst of the sacrum, not previously reported to occur in this area, caused radicu-

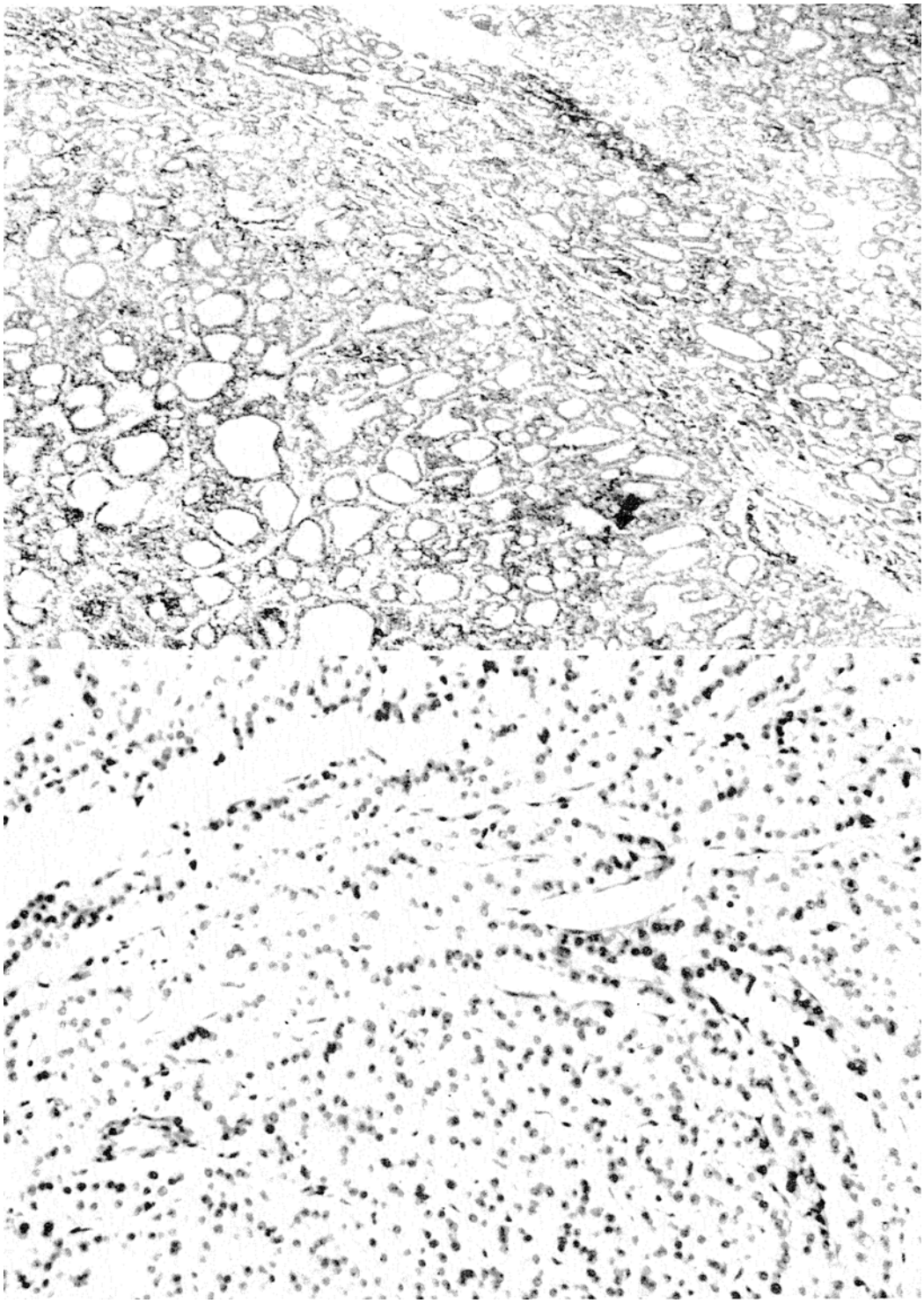

FIG. 7 *(above).* Presacral teratocarcinoma with a one-sided development toward a thyroid follicular carcinoma (×85, reduced from ×100).

FIG. 8 *(below).* Presacral adrenal rest tumor (×170, reduced from ×200).

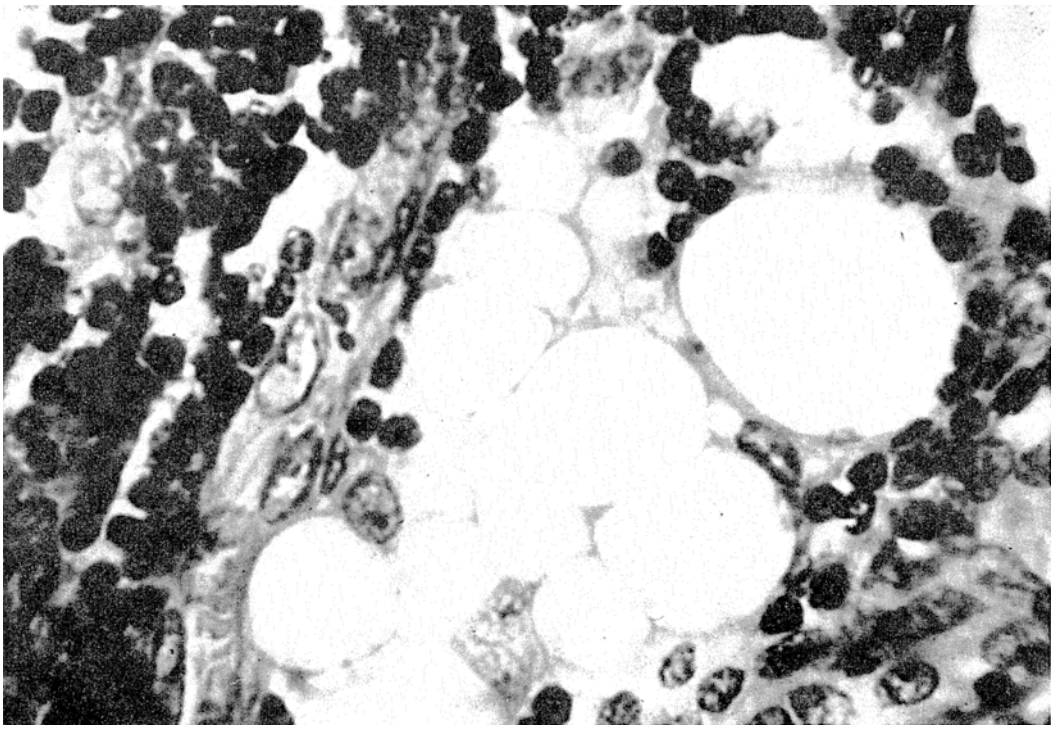

FIG. 9. Presacral lipogranuloma (×340, reduced from ×400).

lar pain in the lower extremities of a 50-year-old woman. X-rays revealed a large central defect in the third sacral segment. Removal confirmed the diagnosis.

The osteogenic sarcoma was found in a 26-year-old white man, who rapidly succumbed to his tumor.

The patient with the osteoma of the sacrum was a 20-year-old woman. The tumor had its onset during her first pregnancy, with pain and weakness of the lower extremities. Results of pelvic and rectal examinations at that time were reported "negative." Because of continued complaints, self-hypnosis was advised, but was not successful. An x-ray two months later demonstrated an "incidental grapefruit-sized mass in the sacrum," confirmed by pelvic examination as close to the rectum, and firmly fixed to the sacrum. Incisional biopsy gave a tissue diagnosis of osteoma. The patient was subsequently delivered of a term pregnancy by cesarean section. In the ensuing four years, she delivered two other term pregnancies via cesarean section. No further follow-up data could be obtained.

Miscellaneous

The miscellaneous group of patients had an average age of 61 years. Sixteen cases represented 25 per cent of the series (Table 1). There were nine locally persistent and metastatic carcinomas, two liposarcomas, one hemangioendothelial sarcoma, one malignant tumor (type undetermined), one plasma-cell myeloma, a lymphangioma, and an extra-abdominal desmoid tumor. The last two lesions have not previously been reported in the English literature. The two liposarcomas were found as presacral masses in women 76 and 83 years of age. One had been treated with radiation therapy for carcinoma of the uterus years before. Both died rapidly of their disease.

TABLE 4. *Neurogenic Lesions*

Tumor	Patient's Age (Years), Sex	Presenting Complaint	Physical Examination	Surgical Procedure	Follow-up
Neurofibroma	28, F	Chronic severe back pain and right buttock and rectal pain; examination by orthopedic and neurosurgical services negative; referred to radiating to right	Mass found on digital examination with patient in standing position performing Valsalva maneuver	Trans-abdominal excision	2 years No recurrence
Neurofibrosarcoma	42, M	psychiatric service Pain in tailbone, thigh	Mass found on rectal examination (small mass)	Kraske	11 years No recurrence
Neurolemmoma	17, M	Weakness in right leg and rectal pain	Fullness, right buttock; weakness, right leg; large presacral mass (13×7×8 cm)	Kraske	3 years No recurrence
Ependymoma	29, M	Back pain, leg weakness	Patient had had first operation 13 years before for an ependymoma of the dorsal spine, now had recurrence in presacral area (massive)	Coccygectomy and sacral laminectomy	Recurred
Ganglioneuroma	26, F	Backache	Mass had interfered with pregnancy 3 years earlier; removed fourth month of second pregnancy (3.5×2.1×.5 cm)	Kraske	14 years No recurrence
Ganglioneuroma	46, M	Asymptomatic	1-cm mass found on routine physical examination	Trans-abdominal excision	18 years No recurrence

The hemangioendothelial sarcoma developed in a 35-year-old Catholic nun who complained of low back pain which radiated to the left leg. No pelvic or rectal examination was reported on the hospital record. Continuing pain prompted further x-rays, including a myelogram. Conservative therapy did not help. Subsequent sacral laminectomy revealed the tumor. The patient died a year later of widely disseminated disease.

The lymphangioma was discovered during a prepartum examination (Fig. 10). An elective cesarean section was done. A neurosurgeon was consulted in the operating room, and a subtotal removal of the multilobulated soft cystic mass was accomplished. No recurrence has been found in three years.

The extra-abdominal desmoid tumor was found in a 54-year-old white woman who complained of rectal discomfort. The mass was approached transperineally, and was found to be trilobed, with a large component in the presacral space. The other two components were lateral to the rectum. This proved to be an extra-abdominal desmoid tumor. Two years postoperatively, there is no evidence of recurrence (Fig. 11).

There were nine patients with locally persistent or metastatic carcinomas in the series (Table 5). Their average age was 60 years. These cases are included in the series because they were diagnostic problems, manifesting as presacral masses many months to years after initial resection of the primary tumors. In one case, the presacral

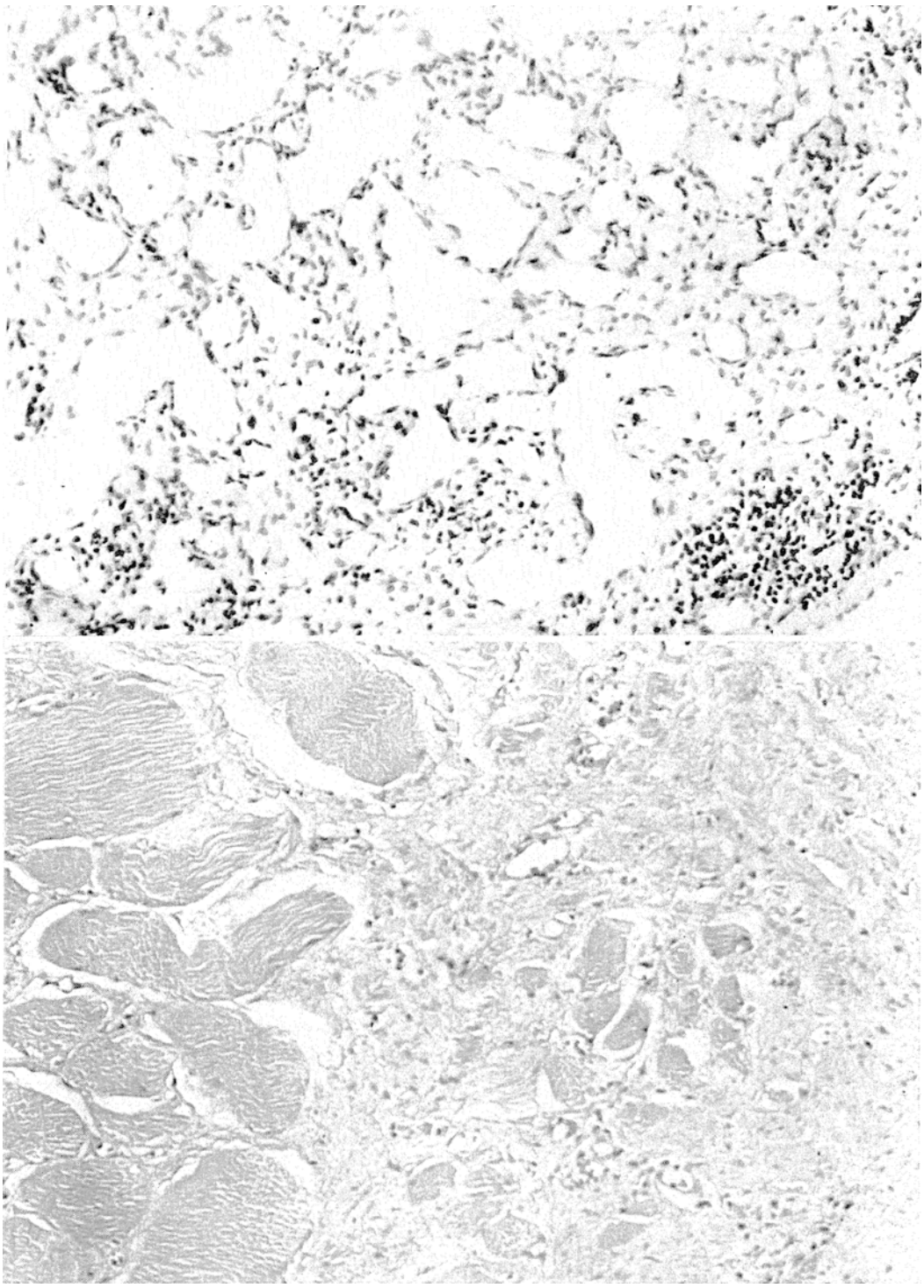

FIG. 10 *(above).* Presacral lymphangioma (×170, reduced from ×200).

FIG. 11 *(below).* Presacral desmoid tumor (×85, reduced from ×100).

TABLE 5. *Locally Persistent or Metastatic Carcinomas—Nine Cases*

Primary Tumor and Therapy	Patient's Age (Years), Sex	Tumor-free Interval	Presenting Complaint	Physical Examination	Follow-up Remarks
1) Carcinoma of the rectum Abdominoperineal resection	65, F	4 years	Ache in the anal area	6–7-cm smooth, hard presacral mass	Removed through a Kraske approach No recurrence after 10 years
2) Carcinoma of the rectum Abdominoperineal resection	72, F	6 years	Increasing severe pelvic and sacral pain	Large presacral mass firmly attached to the sacrum	Transperineal needle biopsy of mass—adenocarcinoma; died 7 years after primary surgery of metastatic disease
3) Carcinoma of the rectum Abdominoperineal resection	39, F	3 years	Hitting something with douche nozzle	6-cm fixed mass in the presacral space	Incomplete resection of metastatic carcinoma of the rectum involving the sacrum; died 4 years after primary lesion was resected
4) Carcinoma of the rectum Abdominoperineal resection	74, F	1 year	Perineal pain	4-cm tumor in the presacral space	Needle biopsy—metastatic tumor; radiation therapy; no follow-up
5) Carcinoma of the rectum Abdominoperineal resection	51, M	3 years	Drainage from perineal area Multiple recurrent abscesses	Pain to palpation in the peritoneum	Methylene blue dye injected into sinus tract and Kraske approach demonstrated presacral lesion; sinus tract was curetted, wound packed and microscopic examination revealed metastatic carcinoma with invasion of the coccyx; no follow-up
6) Carcinoma of the rectum Abdominoperineal resection	50, M	6 years	Perineal pain	2-cm palpable mass anterior to the coccyx and fixed	Kraske approach; microscopic examination revealed metastatic tumor involving the coccyx; no follow-up
7) Carcinoma of the bladder Radical cystectomy	59, M	5 years	Swelling of left leg	Presacral mass	Needle biopsy—anaplastic carcinoma; died of disease
8) Carcinoma of the prostate Retropubic prostatectomy	67, M	4 years	Rectal discomfort	Large presacral mass	Needle biopsy—metastatic carcinoma of the prostate; bilateral orchidectomy; died of disease
9) Carcinoma of the kidney	59, M	Patient had difficulty in urination. A "normal" C & P was obtained. TURP was performed for BPH. Three months later he complained of low back pain. X-ray showed osteolytic lesion in one sacral segment. Rectal exam revealed large presacral mass; biopsied two times and reported to be a "chordoma." Sacral laminectomy obtained tissue consistent with the "chordoma." Repeat IVP revealed a lesion of the left kidney. Nephrectomy obtained a clear-cell carcinoma, identical to the presacral metastases. Patient died 5 months later of pulmonary spread.			

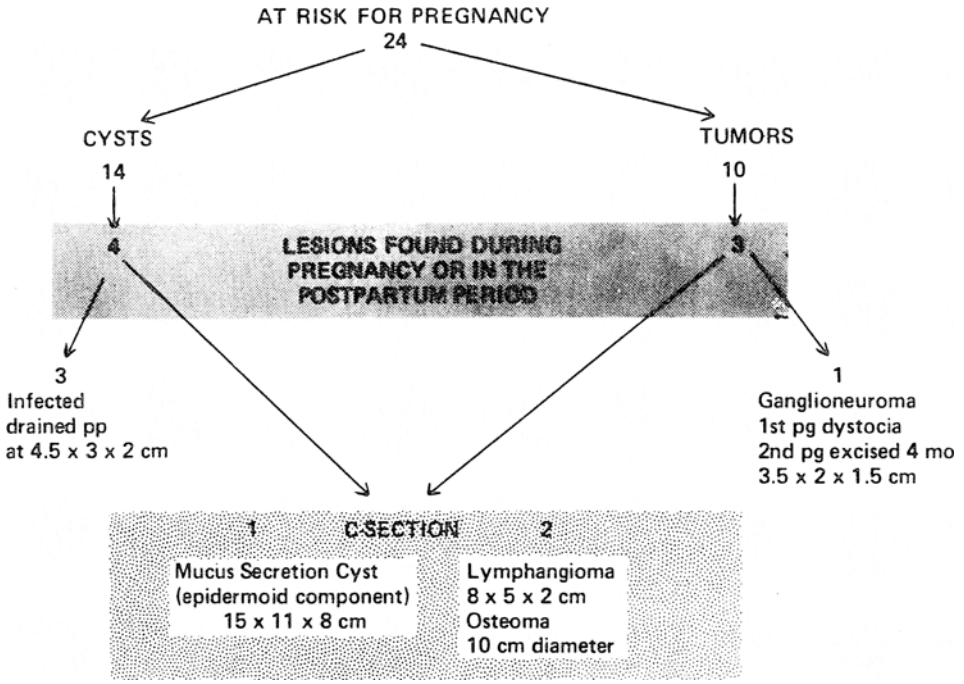

FIG. 12. Presacral lesions found during pregnancy or in the postpartum period.

metastatic tumor developed before the occult primary was discovered. Overall, as might be expected, there was an especially dismal prognosis for patients in this category.

Malignancy

The chance that a presacral lesion is malignant is approximately 40 per cent (26 of 63 patients), considering the entire group of collected cases. It would appear that a presacral mass is more likely to be malignant in a male patient. Nine of 17 male patients, or 53 per cent, had malignant tumors, and seven of them (77 per cent) died with their tumors. Seventeen of 46 female patients, or 37 per cent, had malignant masses, and 11 of these patients, or 64 per cent, died with their tumors.

However, if the nine patients listed in Table 5 with locally persistent or metastatic lesions are deleted because of the information gained from their histories, then the incidences of malignancy in male versus female patients are almost identical: four of 12 men, or 33 per cent, and 13 of 42 women, or 31 per cent. Conversely, two of three patients, male or female, can be expected to have a benign cyst or tumor, but if the mass is malignant, then the prognosis is grave for either sex.

Pregnancy

Twenty-four women were at risk for pregnancy in this series. Seven women were either pregnant or in the postpartum period when their lesions were discovered (Fig. 12).

One of the patients, who had a mucus-

secreting cyst containing a large epidermoid component, required cesarean section; three other women with smaller cysts became symptomatic in the postpartum period because of rectal drainage and infection.

Three women noticed the onset of presacral tumors during pregnancy. Two required cesarean section; one for a lymphangioma, previously mentioned, and the other for an osteoma of the sacrum. The third woman in this group had a ganglioneuroma that caused dystocia during her first labor. The tumor was not excised until the fourth month of her second pregnancy, from which she subsequently was delivered of a viable infant.

Discussion

Various estimates of the relative incidences of presacral cysts and tumors have been attempted.[3, 13, 18, 22, 25, 27] McColl[19] could find only 23 of these lesions in the records of St. Mark's Hospital. Jackman and Clark[9] reported 82 determinate cases from the records of the Mayo Clinic over a 14-year-period. Suffice it to say, these lesions are rare.

When the findings of our series were compared with other series, a number of discrepancies became apparent.[3, 18, 25] Our study was limited to the adult population. Most previous reports present their total pediatric and adult experience with these lesions. It is important to remember that teratomas and meningoceles are usually found in female infants and children.[13]

Some centers recorded a large number of chordomas and a paucity of developmental congenital cysts.[3, 9, 19] Freier *et al.*,[3] from the University of Michigan Medical Center, in fact, recorded finding no mucus-secreting cyst. We submit, therefore, that our series represents a more accurate relative frequency of distribution of presacral lesions in adults in the general population.

Two features are outstanding when the symptom complex of presacral lesions is studied; many of these patients complained of pain while sitting, and many others developed persistent symptoms after falling on their buttocks. Generally, the symptoms seemed to be a function of the size and location of a lesion, and the presence or absence of infection. A patient with abdominal pain and a pelvic mass enlarging superiorly might come to the attention of a general surgeon or a gynecologist. A mass enlarging anteriorly compressing the colon and bladder and displacing the uterus might engage the attention of a urologist or obstetrician. These lesions may interfere with vaginal delivery of a term pregnancy. Infected cysts presenting inferiorly may masquerade as a fistula, abscess, or other anorectal problem, and thus gain the attention of a colorectal surgeon. Several patients in this series were seen because of low back pain, sometimes with radicular compression symptoms, and as a consequence, were initially examined by an orthopedic surgeon or neurosurgeon.

The diagnosis of a presacral lesion usually can be made by doing a careful rectal examination. Many of these lesions are soft, compressible, and difficult to appreciate. It is necessary to rotate the examining finger around to feel the presacral space. One should also look for an anal dimple or sinus tract. The lesions of several patients were actually misdiagnosed and inadequately treated for years before the correct diagnosis was made and definitive surgery performed. Other aids to diagnosis are proctoscopy and various radiographic procedures, including plain films, barium-enema studies, intravenous pyelogram, sinugram, and occasionally, arteriogram.

The Kraske‡ procedure (Fig. 13)[12, 16, 24] and the transperineal route were the most frequently employed surgical approaches.

‡ Paul Kraske was a German surgeon who first described his approach to the presacral space in 1885 as a method of resecting rectal carcinomas. Through a posterior midline incision, the coccyx is excised, gaining exposure of the presacral space.

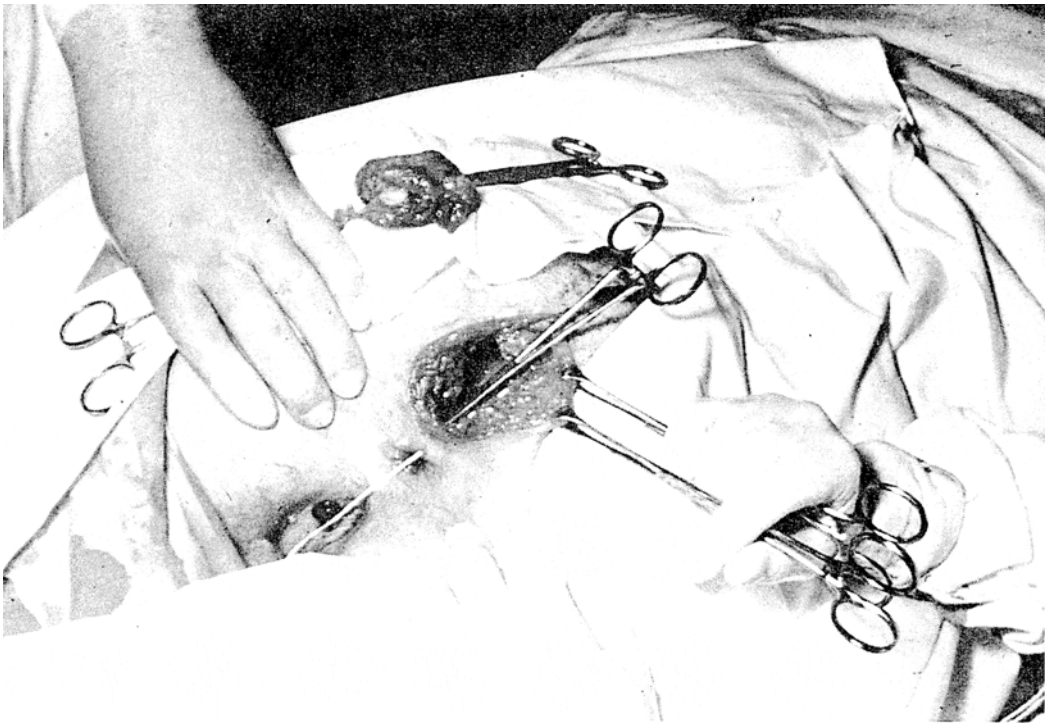

FIG. 13. Presacral developmental cyst (specimen on left buttock). Removed by Kraske procedure. Note probe in postanal sinus tract immediately dorsal to anus. (Courtesy of Donald R. Laird, M.D.)

Both were very satisfactory in all cases where properly applied. Nine times, deep pelvic lesions were approached through the abdomen, and in four instances, this approach was unsatisfactory because of inability to excise the mass completely. One planned abdominoperineal procedure was successful in the excision of a large epidermoid cyst filling the pelvis. The other surgical needle biopsy, were used for diagnosis only, maneuvers, including incisional biopsy and particularly in the locally persistent and metastatic carcinoma group.

Conclusions

A study of 63 cases of presacral tumors and cysts in adults is presented. We believe this series represents a reasonably accurate distribution of lesions encountered in this area in the general population.

Seventy-three per cent of the patients in the study were female. All patients were Caucasian except one American Indian. Congenital developmental cysts were found overwhelmingly in middle-aged women. The terminology of these cysts is discussed.

The malignancy rate in either sex is approximately one in three. In both sexes, with a malignant presacral tumor, the prognosis for survival is grave.

The diagnosis depends on doing a careful rectal examination. Tissue diagnosis can be obtained when indicated by doing an incisional or needle biopsy. The Kraske procedure and the transperineal approach were the most satisfactory surgical techniques for excising a small low-lying lesion. When larger lesions that involved multiple systems were encountered, a multi-discipline team approach was much preferred.

Four new lesions of the presacral space are described.

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