# Sacrococcygeal Teratomas in Adults

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Eleven sacrococcygeal teratomas in adults which were surgically treated from 1952 to 1972 were reviewed. All were benign, and all but one occurred in females. Pain and drainage were the most common symptoms, and presacral masses were found in all on rectal examination. An external tumor was present in only one. Four were large, filling the pelvis. Roentgenographic examinations, particularly barium enema and cystogram, were helpful both diagnostically and in determining extent of the lesion. Calcium deposits were present in three, and none revealed bony destruction. All tumors were surgically resected; however, five had one or more preliminary drainage procedures. Nine were resected via the posterior approach, and two large tumors required abdominoperineal resection. There were two recurrences one of which was cured after re-excision and coccygectomy. Management of special problems in complicated cases are discussed, technical maneuvers found helpful are presented and differences between the adult and infantile varieties are evaluated. A review of literature concerning these tumors and controversial aspects of terminology and diagnostic criteria, which could account for the paucity of reports of this lesion in adults, are presented.

**S** ACROCOCCYCEAL TERATOMAS IN INFANTS were known by the ancients, and their occurrence in the newborn were considered by the Chaldeans as harbingers of prosperity in the land.<sup>4</sup> Similarly, in the modern era this condition in children, though not common, is well known, surgical treatment is standardized, results in general are good, but whether it brings prosperity, even to the surgeon, is doubtful.

In adults, however, these tumors are less common and reports are few and isolated. The literature reveals such confusion in terminology and definition<sup>14</sup> that determination of the incidence is virtually impossible. Our review reveals only 24 to have been reported in the past 20 years. However, it is probable that many have been reported under other designations which will be discussed. From the Surgical Service of the Baptist Memorial Hospital and the University of Tennessee College of Medicine, Memphis, Tennessee

This paucity of reports on these tumors in adults prompts us to review the experience at Baptist Memorial Hospital, Memphis, with a total of 17 sacrococcygeal teratomas, 11 of which occurred in adults. Since this entity in infants has been amply documented by others, the report will deal only with the adult variety.

#### **Characterization of Sacrococcygeal Teratomas**

## Incidence

These tumors are rare. Hospital admission incidence varies from one in  $30,000-170,000.^{6,7,9}$  The majority occur in infants<sup>4,9,11,15</sup> with a female preponderance of 4:1.<sup>3</sup> Incidence in the adult is probably unknown. Incidence in twins varies from approximately 10 to  $50\%.^{4,15}$ 

## Origin

They probably originate from the totipotent remnants of the primitive knot in the sacrococcygeal region, but the cause of their occurrence is unknown. Theorists believe the female preponderance may be due to the fact that the ovaries differentiate later (10 weeks) in embryonic life than do the testes (7 weeks).<sup>7</sup>

## Pathology

Diagnostic criteria specify tumors arising from the sacrococcygeal region which contain tissue derived from more than one primitive germ layer.<sup>4,9,10,15</sup> Some investigators believe that the frequent predominance of tissue from a single layer, usually ectoderm, may necessitate a careful microscopic search to rule out tissue from endodermal and mesodermal derivatives and if a careful enough search of presacral cysts is made, that derivatives of all three germ layers can usually be found. For this

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reason, many of these tumors may have been referred to as presacral dermoid or anterior pilonidal cysts, and some cases reported as enterogenous cysts conceivably could be one-sided developments of teratomas. Others<sup>6,14</sup> however, do not believe presacral cysts should be classified as teratomas.

Approximately 85% are benign;<sup>15</sup> however, there is a tendency for malignant change with increasing age, particularly after the age of four months.<sup>7,15</sup>

In infants these tumors are externally visible in about 90%,<sup>10</sup> although they may be intrapelvic. They are usually bulky and are composed of cystic and solid elements. They may contain calcific deposits or teeth which may be visible on X-ray examination and aid in differential diagnosis. Destruction of the coccyx or sacrum usually does not occur unless they are malignant.

Microscopically, in addition to dermal derivatives, almost any type of tissue such as smooth and striated muscle, cartilage, fat, brain, nerves, and intestinal mucosa may be seen. According to Hanley<sup>6</sup> all types of tissue have been found except cardiac muscle and placental tissue.

## Differential Diagnosis

Chordoma, meningocele, neurofibroma, fibrosarcoma, giant cell tumor of sacrum, pilonidal cyst, osteomyelitis of sacrum, fistula with presacral extension and abscess formation, postinjection granuloma, and tuberculosis may be confused with sacrococcygeal teratomas.

Chordomas are malignant, painful, most often seen in adults, and are associated with bony destruction of the sacrum or coccyx.<sup>8</sup> Similarly, giant cell tumors occur more frequently in adults, occasionally are pulsatile, and are associated with destruction of bone.<sup>1</sup> Meningoceles are usually accompanied by spina bifida and are cystic and covered with a translucent membrane. Bony changes are present in osteomyelitis, and presacral cyst or tumor formation is usually absent.

Neurofibromas and fibrosarcomas are usually solid, painful, and may be associated with bony erosion, but they may be impossible to differentiate preoperatively. Pilonidal cysts and sinuses occur posterior to the sacrum and coccyx and are rare in infants, children, and females. Anal fistula with presacral abscess is impossible to differentiate preoperatively from an infected cystic sacrococcygeal teratoma. Oleomas secondary to injection treatment of hemorrhoids may be found in the presacral space and pose a difficult diagnostic problem in the adult.<sup>6</sup>

#### Treatment

Complete surgical excision usually yields good results except in the malignant type.<sup>4,10,15</sup> Incomplete excision and surgical manipulations other than complete excision may increase the likelihood of malignant change.<sup>7</sup> The coccyx should be routinely removed and as much of the sacrum as necessary.

If resection of the third or a higher sacral body is necessary S-2 and S-3 nerves should be transplanted bilaterally and preserved for protection of bladder control.<sup>2</sup> Shackleford<sup>11</sup> and Swinton and Lehman<sup>13</sup> state, however, that preservation of S-3 on one side is sufficient.

preservation of S-3 on one side is sufficient. Smith, Passaro, and Clatworthy<sup>12</sup> have shown in infants that the middle sacral vessels frequently comprise the only significant blood supply to these tumors. Therefore, they suggest ligation and division of these vessels before tumor resection, if possible, to avoid both troublesome

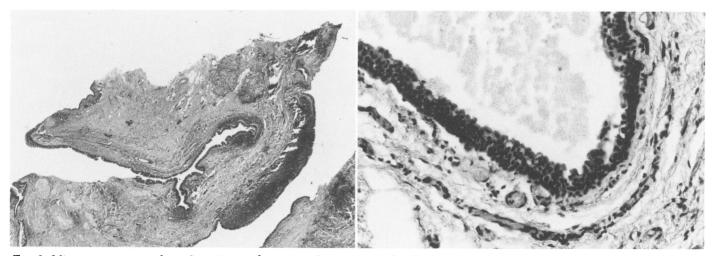


FIG. 1. Microscopic sections from Case 10 revealing tissue derivatives of the three primitive germ layers: (Left) Squamous epithelium (left upper); glandular structures, probably salivary (right upper); pseudostratified columnar epithelium, probably respiratory and lymphoid tissue (right lower), and neural and mesenchymal tissue (left lower). (H. and E.  $\times$  50). (Right) Area of transitional epithelium probably representing the genitourinary tract. (H. and E.  $\times$  250).

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5	55 M			)	+	1					1	I	Posterior	I	I	I	Died Ca stomach 1 yr. postop.
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0	F 56	+	+	1 yr.	+	1	ł	+	1	I	1	+	Posterior	I	I	1	Good
7	F 18	+	I	18 yrs.	+	+	I	+	I	I	I	1	Posterior	I	I	1	Good
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Only 1 week postop.

hemorrhage and tumor dissemination in case of malignancy.

## **Clinical Material**

Diagnostic criteria for inclusion in this report as sacrococcygeal teratomas were patients with tumors arising in the sacrococcygeal region which contained tissue derivatives of two or more primitive germ layers (Fig. 1).

Seventeen such patients were treated at Baptist Hospital from 1952–1972. During the same interval 870,268 patients were admitted, giving a total incidence of approximately one in 54,000 hospital admissions and an adult incidence of approximately one in 87,000. Eleven were in adults varying from 18 to 76 years of age, and all but one of these were in females (Table 1). Six were in infants, only one of whom was a male. Only one was malignant; a non-resectable adenocarcinoma in a 14 months old female infant who died eight months later.

#### Symptoms

Pain and drainage from the sacrococcygeal region were the most common symptoms with pain present in seven and drainage in four patients. Only one patient complained of an external tumor mass. Symptoms had been present in three patients practically all their lives and in four, the duration varied from one to two years. Four were completely asymptomatic with the tumors being discovered on routine physical examination. One of these was pregnant and required a Cesarean section, because of the tumor, six months prior to its resection.

## Physical Findings

Presacral cystic masses were found on rectal examination in all 11 patients. Only one presented an external tumor mass and this involved the left buttock and extended into the presacral space. These masses were huge in four patients and filled the pelvis so tightly in two that the rectum and vagina were occluded. In Case No. 8, digital vaginal and rectal examination was impossible due to the pressure of the mass, the abdomen and bladder were distended, and the urethra was obstructed (Fig. 2). Cystocutaneous, cystorectal, or cystovaginal fistulae were present in three patients and separate pilonidal cysts were found in two.

## X-Ray Findings

Roentgenographic examination revealed no evidence of bone destruction in any patient. The diagnosis of teratoma was suggested by calcification within the tumors in three patients (Fig. 2). Barium enema examinations were performed in six patients and revealed anterior displacement of the rectum in five (Figs. 3, 4). Lipiodol injection of a fistulous tract revealed communication with the rec-



FIG. 2. Scout film of abdomen in Case 8 showing pelvic mass containing area of calcification.

tum in one patient. In another, accurate localization of the cystic mass was determined by injection of Cystokon 15% into the cyst through a previously placed catheter (Fig. 5). In this patient intravenous pyelogram revealed medial displacement of the left ureter by the tumor.

#### Laboratory Findings

Laboratory determinations generally were not helpful diagnostically. Two patients exhibited leucocytosis and both had perforation with fistula formation.

#### Pathologic Findings

Gross. All 11 tumors were cystic and located in the presacral space. They were multilocular in four patients, and two separate cysts were present in two. In Case 11 there were both presacral and left gluteal components, and many separate cysts honeycombed the adjacent tissues. Separate pilonidal cysts were present also in two patients.

In most cases these cysts were markedly adherent to the coccyx and rectum, and in cases with fistula formation, clevage planes were virtually nonexistent. Cysts usually contained thick yellow-white grumous material and hair was present in three (Fig. 6).

*Microscopic*. All these neoplasms revealed derivatives of more than one germ layer, and a variety of different structures were seen, including squamous epithelium, nerves, central nervous tissue, hair, smooth and striated

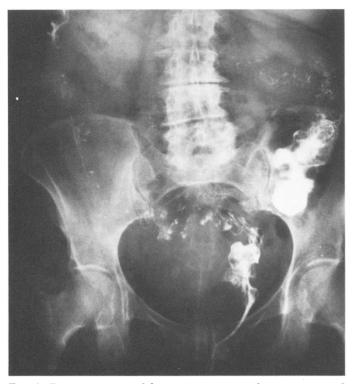


FIG. 3. Roentgenogram of barium enema revealing anterior and lateral displacement of rectum by presacral tumor in Case 8.

muscle, intestinal mucosa, respiratory epithelium, cartilage, glandular structures, and transitional epithelium.

## **Operative Procedures**

Complete resection was performed in all patients with coccygectomy included in ten. Two required partial removal of the sacrum. Preliminary drainage procedures had been performed prior to admission or were necessary prior to resection in five patients. Two required diversionary colostomies before resection. A second resection was necessary for recurrence in two patients, one of whom had previous resection without coccygectomy. Two patients required two, two required four, and one patient required five operations before cure was obtained.

The following standard procedure was performed with minor variations in nine of the 11 patients: the patient was placed in the prone jack-knife position and a posterior midline incision was made from the mid-sacrum to beyond the tip of the coccyx. The posterior surface of the sacrum and coccyx were bared, the anococcygeal ligament was cut and muscular attachments to the coccyx were divided. The coccyx was disarticulated and either removed or left attached to the cyst. The presacral space was explored, and the extent of the tumor determined. In two patients a portion of the sacrum was removed with the Gigli saw after dividing the ligamentous and muscular attachments. The middle and lateral sacral vessels were ligated, if possible, and divided. The cyst was then dissected out taking care to avoid perforation of the rectum. Hemostasis was secured, and the wound was closed with drainage. Drains were removed in five to six days, and the patient was ambulated about one week postoperatively.

Two patients required abdominoperineal procedures. In one, the tumor was freed up as far as possible from above, then removed via the perineum through a "U" shaped incision with preservation of the rectum. In the other, abdominal exploration revealed the tumor to be too low for dissection from above, and it was removed from below.

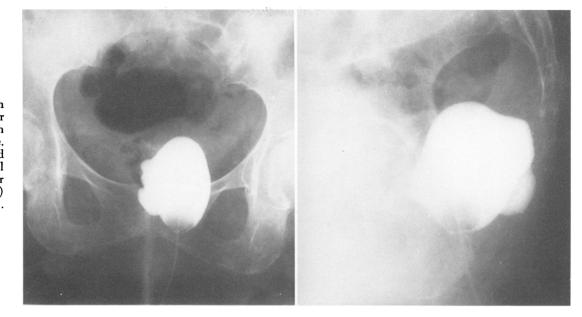
In addition to preoperative antibiotics, mechanical and chemical bowel preparation were used in the majority.

Special problems were encountered in several patients: Case 3 was complicated by both cutaneous and rectal fistulas, and dissection was difficult due to inflammation and fibrosis. This lesion recurred after initial resection, and a diversionary colostomy was necessary. Following this, a second resection was performed which was curative. Fistulagram was helpful in determining the extent of the cyst and detecting location of the rectal fistula. Coccygectomy was not included originally, but was performed at subsequent resection. Because of the



Fig. 4. Anterior displacement of barium-filled rectum in Case 10 showing extension of tumor to level of second sacral body.

FIG. 5. Cystogram through Foley catheter placed in cyst through ischiorectal drainage site. This reveals marked shrinkage from original size (Fig. 2 and 3) after drainage. PA view (left) and lateral view (right).



inflammatory reaction and infection, primary closure was deemed unwise, and the wound was packed open. Healing required approximately one year, but the result has been good.

Case 8 was the largest and most difficult to manage. This 74-year-old white female presented with a huge tumor tightly filling the pelvis, obliterating the vagina and rectum, and obstructing the urethra. Spontaneous transvaginal perforation occurred shortly after admission, and symptoms gradually subsided. Past history revealed an abdominovaginal resection of a dermoid cyst 30 years previously. Subsequently, two additional transvaginal and ischiorectal drainage procedures were necessitated before final resection. The cystic tumor, which was originally huge, shrank so tremendously after drainage that it could be removed posteriorly. Preoperative cystogram through the ischiorectal drainage site was helpful in determining the extent of the lesion and saved the patient an abdominal exploration. Alternative filling and emptying the cyst with saline through an indwelling purse-stringed Foley catheter was of inestimable value in delineation during resection, which was difficult because of absence of clevage planes and inflammatory reaction. In two areas the cyst wall was inseparable from the rectum, and small portions of the rectal wall were resected with the cyst. These defects were closed in two layers without complications. The patient recovered uneventfully and has been well for five years.

Case 10 presented with a tumor which extended almost to S-1 and required removal of the lower one-third of the sacrum. Dissection was extremely difficult because of fibrosis and subacute inflammation, and it was inadvertently entered during dissection. It was necessary to open the cavity widely, aspirate the contents, and utilize the finger inside the cyst for traction and tension to permit resection without perforation of the rectum.

Case 11, despite absence of symptoms, presented an extensive lesion which consisted of both intrapelvic and external components. Only the presacral mass was detectable on examination. Exploration revealed a mass with multiple extensions about the rectum and one large hour-glass component which surfaced through the left Gluteus maximus muscle onto the left buttock. There were virtually no cleavage planes, and it was necessary in this case also to open the cyst widely to determine its true extent and trace out the many projections, which extended in spider-leg fashion around both sides of the rectum and into the buttock. In addition to the central cystic portion the precoccygeal tissues were honeycombed with separate small cysts which also involved portions of the levator and external sphincter muscles, which were resected.

## Complications

Complications were few and of minimal degree. Delayed wound healing was present in two, in one of whom the wound was packed open because of infection. In the other an inverted "U" incision had been used. Blood was required in only two patients. One of these had an abdominoperineal procedure, and in the other the tumor was resected with the patient in the lithotomy position and troublesome oozing was encountered. This was controlled by temporary packing, and the subsequent result was good.

## Results

Results were excellent in nine patients one week to eleven years after resection. Two developed recurrence

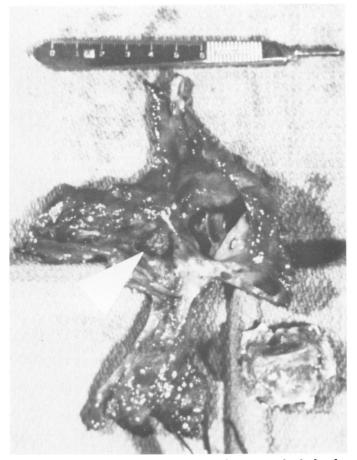


FIG. 6. Opened specimen from Case 10 showing multiple loculations and hair ball (arrow). Towel Clip is in attached coccyx. Portion of sacrum also resected is seen in lower right.

and required a second resection, following which one was cured. The result in the other patient is not known. Case 11 was doing well one week postoperatively at the time of this writing.

#### Discussion

Whether presacral and dermoid cysts in the adult should be classified as sacrococcygeal teratomas is controversial, and, for this reason, their incidence is difficult to determine. We favor their inclusion as sacrococcygeal teratomas if derivatives of more than one primitive germ layer can be found; this was ascertained in the 11 cases reported above. Interestingly, Waldhausen and associates included in their collected series 12 cases reported by others as dermoids. On the other hand, Guillermo and Grossman included 44 dermoids in a total of 80 presacral cysts collected up to 1972. Excluding these, only 24 sacrococcygeal teratomas in adults could be found in a review of the English literature since 1952.

This study suggests the diagnosis be considered in any patient complaining of pain in the sacrococcygeal region, with or without a history of drainage from this area, in whom a presacral mass can be palpated. The value of including the rectal examination as a part of routine physical examination is evident in that one-third of these cases were discovered on routine physical examination in asymptomatic patients.

Roentgenographic examination is not only helpful in diagnosis by virtue of revealing calcification and the integrity of the sacrum and coccyx, but also in determining the extent of the lesion by cystogram, barium enema, and occasionally intravenous pyelogram. Cystogram was employed only when a fistula was present. The advisability of this in the intact tumor is questionable, because of the possibility of malignancy and danger of tumor dissemination. Arteriography as suggested by Smith and his colleagues may be helpful.

The following differences between the adult and infantile varieties were noted: 10 of the 11 tumors in this series were intrapelvic, whereas in the infant 90% are external. All adult tumors were cystic, while in infants they are bulky and contain solid as well as cystic components. In these adults, all tumors were benign and in the 24 collected cases only one was malignant. In infants the incidence of malignancy is much higher.

Resection via the posterior approach with the patient in the jack-knife position proved satisfactory for uncomplicated medium and small-sized tumors.

Patients with huge tumors and those in whom perforation and fistula formation had occurred, or in whom previous surgery had been performed, presented special problems. The value of preliminary drainage in the huge pelvis-filling variety and in those with infection or perforation is shown in Case 8. This tremendous cyst shrank so much after drainage that it was possible to remove it by the posterior route. In cases where this is not feasible however, the abdominoperineal procedure should be employed, and, in the light of Smith's report, the middle sacral vessels should be secured initially during the abdominal portion of the procedure.

The following technical manuevers were helpful: aspiration to reduce size and facilitate exposure, alternately emptying and filling the cyst with saline for delineation, opening the cyst and its extensions widely to determine extent and "resection from within," and the utilization of a gloved finger of the operator or an assistant in the rectum for delineation of the cyst from the rectum.

The coccyx should be removed routinely, not only for exposure, but to remove the nidus which may contain totipotent cellular remnants and cause recurrence. Recurrence was observed in one case in which the coccyx was not removed.

Resection of enough sacrum to permit adequate exposure and safe dissection should be performed. It is unlikely that resection of the sacrum high enough to pose problems with S-2 and S-3 nerves will be required in dealing with these lesions unless they are malignant, since the larger benign cysts can be removed by com-

bined abdominoperineal procedures without major sacral resection.

Colostomy is usually not necessary, but may be required in cases complicated by perforation or fistula.

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#### DISCUSSION

DR. RICHARD T. SHACKELFORD (Baltimore): I should say that I have not had any appreciable experience with sacrococcygeal teratomas, but have had an experience with some 15 cases of sacrococcygeal chordoma. I forget whether any of the teratomas in Dr. Miles' series were malignant. My experience with teratomas has been almost exclusively in children, where malignancy has been fairly common.

I have little to add to the discussion, but there are some points in the technique we have used for chordomas which are applicable and worth considering for teratomas. The technique was devised by John Waugh and his associates at the Mayo Clinic. Actually, my initial paper before this society, when I was made a member in 1953, was on this particular subject.

I will show two or three slides that emphasize the points that Dr. Miles has beautifully and clearly shown and will demonstrate some other slides.

(Slide) The incision is made in the posterior midline from just above the anus as far up on the sacrum as desired, as shown by Dr. Miles.

(Slide) Then, following development of the lateral flaps, the ischio-rectal fossae are opened bilaterally, also as he showed. Before further dissection a very painful maneuver is to insert a sponge-stick clasping a sponge through the anus into the rectum. By manipulating the intraluminal sponge one can feel where the rectum is and avoid its inadvertent injury. I don't remember our having injured any.

(Slide) After exposing the cyst it may be found necessary to remove a considerable portion of the lower sacrum to obtain adequate exposure for excising the cyst. This can be done with a chisel by chiseling off the spinous processes of the lower portion of the sacrum. This unroofs the spinal canal.

(Slide) Unroofing the canal exposes the sacral nerves which can be seen coursing to their specific foramina through which they exit from the canal. S-4 and S-5 bilaterally and S-3 on one side only can be sacrificed without sphincter disturbance. Some years ago Lloyd Lewis and Orthella Langworthy, at Johns Hopkins, showed that if S-3 is preserved on at least one side, along with S-2 and S-1 bilaterally, sphincter control is preserved. Hence if the sacrum must be transected at the level of S-3 or higher, S-3 bilaterally or unilaterally, and S-2 and S-1 bilaterally can be transplanted intact upward by unroofing their foramina with a chisel, carefully avoiding injury to the nerve. With a nerve hook, each nerve can be gently lifted out of its foramen and transplanted to a level above the level at which the sacrum will be transected and still preserve continence. In the two cases in which we have transplanted S-3 we have had no difficulty with continence. In

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this way the sacrum can be removed higher and provide better exposure for removing the tumor.

(Slide) This shows the foramina unroofed so the spinal nerves can be lifted and transplanted to a higher level.

DR. RICHARD G. MARTIN (Houston): I was intrigued by the title of this paper, because I had not been aware of the nomenclature of sacrococcygeal teratomas in adults. We have had them in children—infants—but not in adults, and I called our pathologist last night to see whether I had missed, or what—and I think that it's just a matter of terminology, as Dr. Miles pointed outas to what you call a true sacrococcygeal teratoma.

The posterior approach is good for small lesions, and those in which you don't have to do an AP resection. I wish he would comment a little on his technique in doing the AP resection in these cases.

We have found in large presacral lesions such as chordomas, neurofibrosarcomas, and the like, that sometimes an anterior approach with the patient in extreme Trendelenburg is a good method with a two-team approach. In doing this, you have to place the patient with the buttocks off the end of the table in the lithotomy position, and then put in severe Trendelenburg when you are ready for the peritoneal part.

Now, when we first did this, we had to use shoulder braces, and in doing so we had one case of a 6-foot, 4-inch, 200-pound basketball player who developed bilateral transient brachial involvement, and this, of course, caused us all kinds of anxiety. So now we use what we call the beanbag, which is a sort of a mattress filled with pellets, and can be molded to the contour of the patient and the air evacuated from the pad, and the pad becomes hard, and this will keep him in place without any shoulder stirrups.

DR. ROBERT M. MILES (Closing discussion): With reference to Dr. Shackelford's question about the incidence of malignancy, this is a very interesting thing. In the newborn it varies, but averages about 15% until the age of four months, and Waldhausen and others have shown that from four months to about 15 years this may increase to 40 to 50 percent. However, in the 24 cases that we collected there were only two instances of malignancy in the adult.

I appreciate Dr. Martin's remarks. With respect to his question about the abdominoperineal procedure, the lesion was simply freed up as much as possible from above, and then resected definitively from below. However, in the light of Smith, Passaro and Clatworthy's work on the blood supply of these tumors in infants, if the abdominoperineal procedure is done the middle sacral vessels should be secured initially. They make the point that this not only prevents troublesome bleeding, but also may prevent tumor cell dissemination in case of malignancy.