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Sacrococcygeal chordoma

A clinicoradiological study of 60 patients

Julius Smith, M.D., M.R.C.P., F.R.C.R. [1], **Robert L. Ludwig, M.D.** [1], **and Ralph C. Marcove, M.D.** [2]

[1] Departments of Medical Imaging and [2] the Orthopedic Service of the Department of Surgery
Memorial Sloan-Kettering Cancer Center, New York, N.Y., USA

Abstract. Sixty patients with sacrococcygeal chordoma, who were seen at this center between 1946 and 1985, were studied with particular attention to the radiographic findings. This study was undertaken because of the large number of these cases and comparison was made between the plain films available in 39 patients and the computed tomography CT studies in 22. Bone destruction was found in 78% on plain films but in 90% on CT. A soft tissue mass was identified in plain films in 60% but in 90% on CT. Calcific debris was found in plain films in 44% but in 87% on CT. Mostly the debris consisted of coarse irregular fragments and probably represented sequestrated necrotic bone. Myelography was performed in only 15 patients. Angiography was studied in 10 cases. Of the 60 patients 88% underwent surgical resection. The tumor recurred in 80% and in only 20% was there no evidence of recurrence. Distant metastases occurred in 24% of patients. Fifty percent survived 5 years; 28% survived 10 years; mean survival 7.5 years.

Key words: Chordoma – Sacrum – CT treatment

Chordoma is an uncommon slow-growing malignant tumor of bone which is virtually confined to the axial skeleton. Outside the spine and skull base, these tumors are extremely rare and may be regarded as curiosities. The tumor is thought to arise from notochord remnants. Normally these remnants are most frequent in the nucleus pulposus, however, ectopic rests of the notochord may be found in the vertebral bodies and sacrum [9]. Most chordomas are thought to arise from these ectopic remnants rather than the nucleus pulposus of the intervertebral disc. Origin from the nucleus pulposus has not been demonstrated.

Although slow growing, a chordoma progresses relentlessly and may reach a huge size, particularly when it takes origin from the sacrum. The tumor usually spreads anteriorly into the capacious pelvic cavity and may only produce symptoms when it reaches relatively massive proportions. The sacrum is by far the commonest site for this tumor and accounts for 50% of all chordomas. The skull base accounts for 35% and the spine for 15%.

Chordoma is very uncommon with considerable variation in the statistics reported from various centers. The commonly quoted figure of 4% [4] of all malignant tumors of bone appears to be too high. A figure of 1% [13, 21] from two other large centers is more realistic.

There is considerable variation in the age of incidence of these tumors depending upon site. In the sacral lesions, the peak age incidence is in the sixth and seventh decades, although it can occur in much younger individuals. In the skull and spine, the peak incidence is at least 10 years younger – fourth and fifth decade. Children may be affected but chordoma is usually limited at this age to the skull base and cervical spine. It is of interest, however, that the first sacrococcygeal tumor described was found in a 7-month stillbirth [7]. In the sacrococcygeal region the male-female ratio is 2:1. In the other anatomical areas the male-female ratio is usually equal.

A review of all primary neoplasms of the sacrum at the Memorial Sloan-Kettering Cancer Center over a 40 year period revealed 138 patients. Chordoma accounted for over 40% of these tumors. The large number of patients with chordoma

Address reprint requests to: Julius Smith, M.D., Department of Medical Imaging, Memorial Sloan-Kettering Cancer Center, 1275 York Avenue, New York, N.Y. 10021, USA

treated at this Center led us to reevaluate the radiological findings of this tumor as previously documented in the literature [10, 26]. We were particularly interested in studying the value of computed tomography using modern high resolution scanners. Magnetic resonance imaging, only recently available, was also used in one patient. We also reviewed the clinical aspects particularly symptomatology, treatment, and results.

Materials and methods

We reviewed the charts of all patients classified as sacrococcygeal chordoma who were seen and treated at this Center between 1946 and 1985. The patients were identified from the medical records and the bone tumor registry. All were histologically proven. We excluded all patients in whom the clinical data were no longer available. Many of the patients in this study were included in previous studies by Higinbotham et al. [8], Sundaresan et al. [25], and Krol et al. [14]. A total of 60 patients were considered to be acceptable. Eighteen patients in this study were recent and have not been previously reported. Available for review were plain films in 39 patients, myelograms in 15, arteriograms in 10, and CT scans in 22 patients. Most of the CT scans were performed on modern scanners, 15 of these were in patients who had had no prior treatment and 7 were in patients who had recurrent tumors referred here for further treatment. Radionuclide scans were performed in five patients and one underwent magnetic resonance imaging.

Results

Clinical

Patients ranged in age from 26 to 84 years at the time of diagnosis with a median age of 55 years. The male to female ratio was 1.6:1. The most frequent presenting symptom was pain localized to the low back, sacrum, or coccyx and was recorded in 71% of cases. Twenty-three percent had neurologic complaints such as leg numbness, radicular pain, or bowel/bladder incontinence. Nineteen percent were constipated and 13% of patients noted a sacral mass. The median duration of symptoms prior to diagnosis was 12 months with a range from 1 month to 5 years. The patients' pain and neurologic complaints were ascribed to herniated lumbar disc disease in several cases, and it was only the myelogram that eventually led to the correct diagnosis of chordoma.

Radiological (Tables 1, 2)

Bone destruction (Figs. 1, 2, 3)

Plain films. This was found in 78% of untreated patients and in 75% of recurrent tumors on the *plain* films. The margins were usually clearly outlined but were truly sclerotic in only 43% of the untreated patients available for review. Expansile margins were quite uncommon in this study being only present in 3%. Pseudo-loculation giving a honeycombed appearance (Fig. 3) was present in 17% of untreated patients and in 13% of the recurrent tumors.

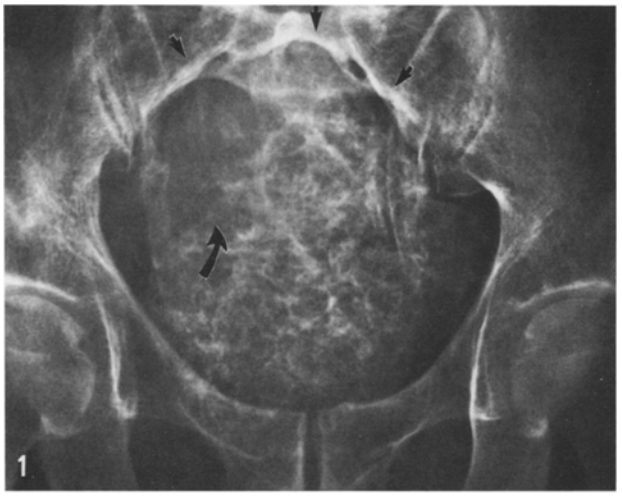
Computed tomography. The CT scan (Figs. 5, 6) not surprisingly gave much more precise informa-

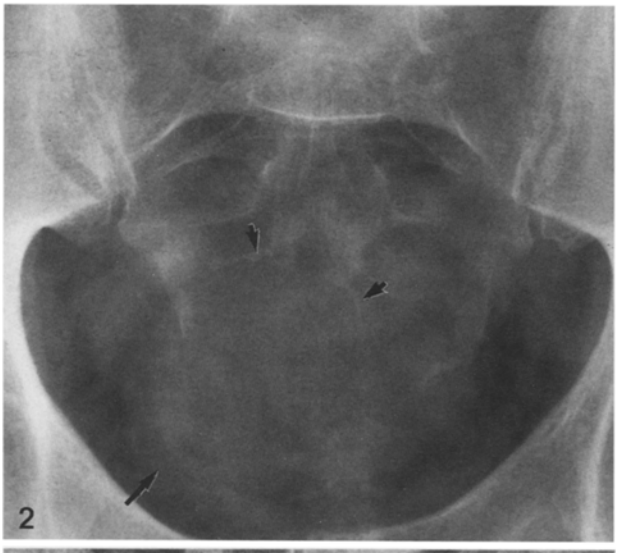
Table 1. Sacrococcygeal chordoma. Plain film findings in 39 patients

	Untreated patients (31)	Recurrent (8)
Bone destruction		
Absent	7 (22%)	2 (25%)
Poorly defined	12 (39%)	4 (50%)
Well defined	12 (39%)	2 (25%)
Sclerotic margins	13 (43%)	1 (13%)
Expansile margins	3 (9%)	–
Pseudo-loculation	5 (17%)	1 (13%)
Soft tissue mass		
Apparent on plain films	18 (57%)	5 (63%)
Calcific debris		
Absent	17 (54%)	5 (63%)
Present	14 (46%)	3 (27%)

Table 2. Sacrococcygeal chordoma. Computed tomographic findings in 22 patients

	Untreated patients (15)	Recurrent (7)
Bone destruction	14/15	6/7
Anterior cortex only	3	–
Posterior cortex only	–	–
Both anterior and posterior cortex	9	5
Sacro-iliac joints	2	4
Soft tissue mass	13	7
Location purely anterior	4	1
Location purely posterior	–	1
Location anterior and posterior	9	5
Calcific debris	14	5
Central pattern	5	3
Peripheral	3	1
Mixed pattern	6	1
Visceral involvement	10	6
Rectum	7	1
Buttock	6	4
Pelvic Muscles	3	3





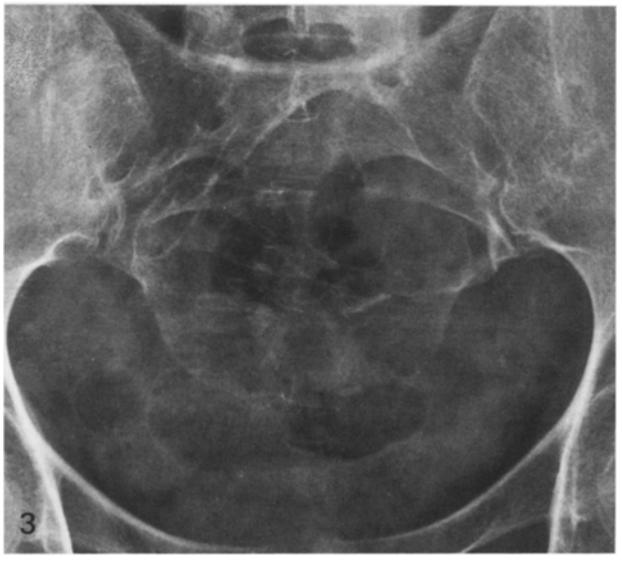


Fig. 1. *Plain film.* Massive tumor involving most of the sacrum which is expanded with a dense superior cortical rim (*arrows*). Extensive calcific debris was present throughout the tumor with a bare area in upper sacrum, where obvious destruction is seen.

tion. In 20 of the 22 patients (90%) destruction was clearly visualized. In previously untreated patients it was 93%. The anterior and posterior cortex were involved in 14/22 patients (64%). Destruction limited to the posterior cortex was not found in any patient.

Soft tissue mass

Plain films. The soft tissue mass was found in only 23/39 of the patients (60%) on plain films. The mass overlaps the bladder and sacrum and may be difficult to recognize. Overlying gas and fecal material may make evaluation extremely difficult.

Computed tomography. On CT examination (Figs. 5–8) the mass when present was demonstrated with ease. It was found in 20/22 (90%) of the cases. The tumors when of any size, and most were large, averging 10 cm, tend to extend forward but also posteriorly into the buttock. This was the case in 14/22 of the cases. In general, the soft tumor mass has a uniform density but in 35% of cases there was irregular lucency indicating necrosis. This was not usually very extensive but in one patient this was of remarkable extent and produced difficulty with the diagnosis. This proved to be a highly anaplastic chordoma with massive necrosis of the tumor.

Calcific debris (Figs. 1, 5, 6, 7, 8)

Material of calcific density was commonly found although on *plain films* in only 17/39 (44%). CT examination, however, demonstrated calcific debris in 19/22 (87%). On CT there are clearly two types of debris that may be found. In a large proportion fragments are large and irregular and may be situated either centrally or peripherally. A second type consists of finer smaller densities. These are usually central. These are less common than the coarse type.

Myelography. The proximity of the spinal theca to the upper sacrum makes it essential that this study be performed in all large lesions involving

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This tumor was unresectable and the patient received palliative irradiation

Fig. 2. *Plain film.* A large area of eccentric destruction in lower sacrum with associated soft tissue mass. Note faintly sclerotic upper margin of tumor (*short arrows*) and soft tissue mass (*long arrows*)

Fig. 3. *Plain film.* Honey-combed appearance due to extensive involvement of sacral foramina. Note sclerotic margins of tumor

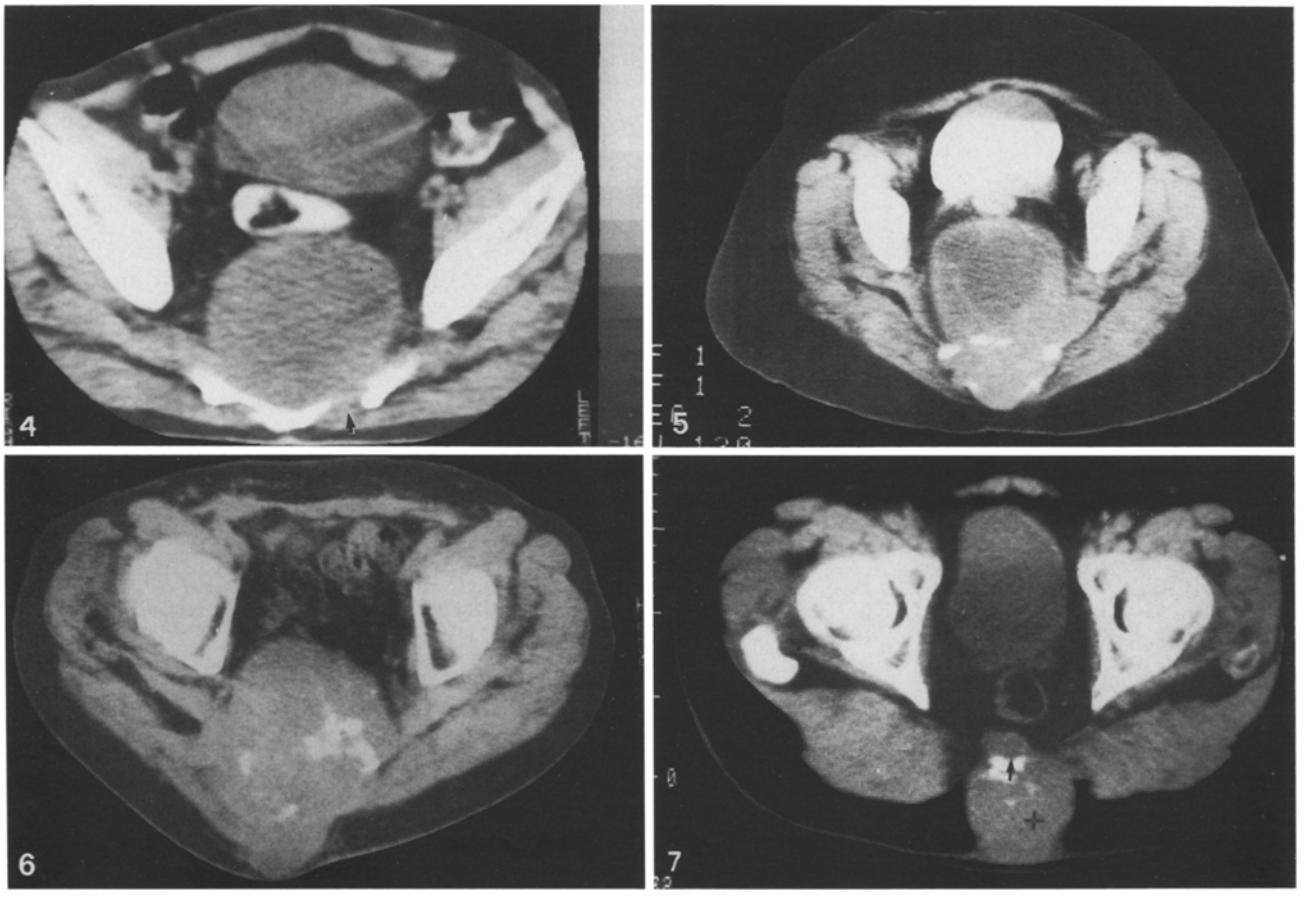


Fig. 4. *CT.* Bulky tumor with small area of destruction involving sacral foramen (*arrow*)

Fig. 5. *CT.* Large tumor with prominent area of necrosis. Tumor extending mainly anteriorly but there is also a small extension posteriorly. Note bony fragments in tumor mass

Fig. 6. *CT.* Bulky tumor extending almost equally anteriorly and posteriorly with involvement of pyriformis and gluteus muscles. Irregular bony fragments are present within the tumor

Fig. 7. *CT.* Soft tissue extends predominantly posteriorly. There is only a small mass anteriorly (*arrow*)

S1 or S2. Even lesions well caudal to this level should undergo myelography since occasionally the soft tissue mass frequently is larger and well beyond the area of bone involvement and may involve the spinal canal. In this study, however, myelography was only performed in a small number of patients probably because so many of the surgical resections were palliative.

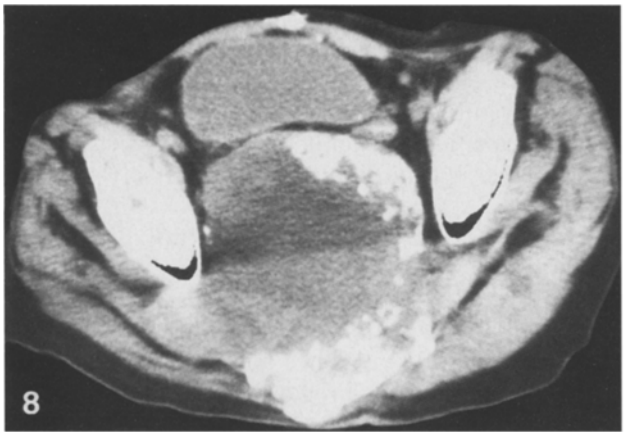
Angiography. Prior to CT this was a relatively common form of investigation for demarcating the extent of the chordoma. By stretching and displacement of vascular structures the size of the tumor could be assessed. Pathological circulation was not demonstrated and despite subtraction techniques, abnormal vascularity was never seen. In this, it is quite different from chordoma of the skull base and spine where abnormal vascularity may be found in as many as 50% of cases [17].

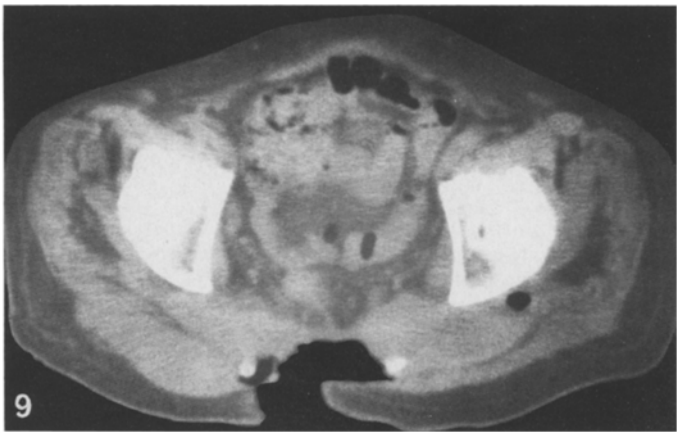
Radio-nuclide scanning. This is only rarely performed but it does produce a pattern which may be specific for this tumor and is helpful in the differential diagnosis of sacral tumors. The main portion of the tumor tends to be "cold" or photopenic and its peripheral margin is surrounded by a halo of increased activity [11]. In four of five patients this pattern was found.

Case reports

Case I

A 43-year-old woman who complained of low back pain for 20 years noted recent exacerbation and sought medical atten-





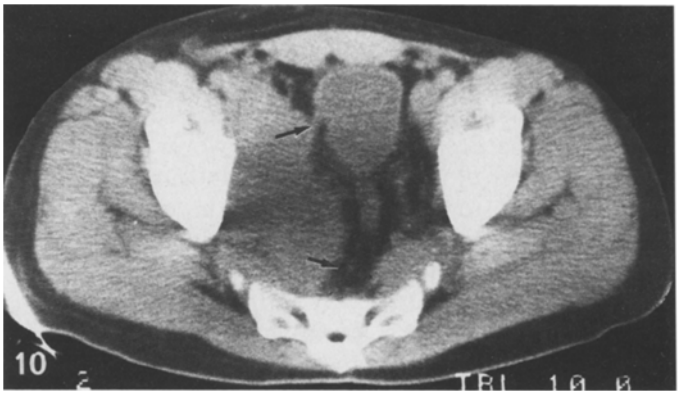


Fig. 8. *CT.* Massive calcific debris localized to lateral aspect of tumor

Fig. 9. *CT.* Deep ulceration through soft tissues at site of bone destruction. Small bony fragments remain. This on biopsy represented a giant and spindle cell sarcoma – a new tumor – almost certainly a radiation-induced sarcoma

Fig. 10. *CT.* Lobulated mass involving side wall of pelvis (*arrows*). Tumor extended into abdomen and led to inferior vena caval thrombus. Patient had a relatively small tumor of the coccyx excised earlier. Tumor recurred in soft tissue only. No evidence of bony recurrence was found

tion. A mass was felt on rectal examination and a barium enema confirmed a retrorectal mass. Plain films of the sacrum showed central destruction in the lower sacrum. She was advised to undergo surgical excision of the mass; however, the patient refused all treatment. Two and a half years later, she returned because of increasing pain, constipation, and abdominal cramps. The area of bone destruction had increased in size. She underwent resection but the margins of the tumor were positive. Two years later she returned with recurrence and a further resection was performed. Three years later a further resection was performed.

Comment. It is seldom possible to observe an untreated early chordoma. While the tumor had progressed, it was still not very large when the patient returned for treatment. CT of the lesion demonstrated that the rate of increase must be regarded as slow. It is probable that if the patient had agreed to surgery initially, the tumor could have been completely resected and a cure resulted. The patient has since had multiple resections and is probably incurable.

Case 2

A 59-year-old man had a sacral chordoma resected elsewhere in 1974. He received 4600 cGy postoperatively. He had persistent low back pain and had two further partial resections. Because of persistent pain, a further course of radiation was given (3000 cGy) in 1978. He was admitted to the Memorial Sloan-Kettering Cancer Center for intractable pain and had a high thoracic chordotomy for pain control.

He developed intractable ulceration of the skin over the sacrum which, when biopsied, revealed a spindle cell sarcoma (Fig. 9). He died 4 months later from pneumonia. At autopsy, there was extensive tumor in the sacrum which showed spindle and giant cell sarcoma. There were also extensive pulmonary metastases of similar etiology. There was no evidence of residual chordoma.

Comment. The second tumor probably represented a radiation-induced tumor. This patient is the only patient with a radiation-induced sarcoma complicating chordoma in our material. There have been only very few reports of postradiation sarcomas associated with chordoma.

Case 3

A 26-year-old man presented with a 3-month history of sacral pain first noted while driving his automobile. Radiographs revealed a coccygeal lesion which was resected at the referring hospital. The patient was referred here for further treatment. He underwent sacral resection of the distal two-thirds of the sacrum and a colostomy was performed. The specimen revealed tumor invading skeletal muscle.

Two months later, he was re-admitted for colostomy closure and 2 months following he developed further recurrence. The CT revealed bulky tumor filling the pelvic cavity and retroperitoneum (Fig. 10). He developed thrombosis of his (R) iliofemoral system. Radiation therapy was given and also chemotherapy. Tumor was encasing the IVC and aorta and extended into the sciatic notch. The patient died 2 years and 2 months following diagnosis.

Comment. This patient was the youngest in the series. His tumor also appeared to be the most aggressive and was totally resistant to treatment. Histologically the tumor was a typical chordoma.

Results of treatment

Of the 60 patients 53 (88%) underwent surgical resection. The extent of disease in 12% was such that surgery was contraindicated. In 80% there was evidence of recurrence and in many cases this occurred several times. In only 20% was there no recurrence documented. This includes long-term survivors and recent patients.

Distant metastases. This occurred in 23.5% of patients. The most common site was the lungs (11%), followed by the skeleton (7%). Metastases to the soft tissues (3.5%) and the liver (2%) were much less common.

Survival. Follow-up was available in 89% of patients. Fifty percent survived 5 years; 28% survived 10 years; mean survived 7.5 years.

Discussion

Sacrococcygeal chordoma accounts for the majority of all axial chordomas, yet it is an uncommon tumor of the skeleton. It does, however, account for over 40% of all sacral tumors in our material and thus has to be considered in the differential diagnosis of sacral tumors particularly in older individuals. It is an unusual tumor, being one of the few that originates from and retains the histological structure of the primitive embryonic tissue. Because of its slow growth and anatomic location, symptoms are frequently late, and at the time of presentation the tumor is very large making curative resection or curative radiation therapy impossible. When a patient presents with a large sacral tumor in which several sacral segments are involved, its exact sacral segment of origin is uncertain. In almost all cases in this study the tumor appeared to arise from the more caudal segments of the sacrum, S3, 4 and 5 or coccyx. None of the tumors appeared to arise from S1 or S2. This is helpful in the differential diagnosis since giant cell tumor, chondrosarcoma, and osteosarcoma are more common in the upper segments. Being generally low in position the tumor is readily palpable on rectal examination. In all patients in this study in whom digital rectal examination was recorded, the tumor was palpated. Unfortunately digital examination is frequently deferred and then forgotten.

Radiolographically the tumor is very characteristic and often pathognomonic. A central area of bone destruction is almost always present and its central position is helpful in the differential diagnosis from other tumors which tend to be eccentric. Rarely the chordoma may be eccentric in position (Fig. 2) and radiological diagnosis then may be difficult. A retrorectal mass containing calcific debris displacing the rectum anteriorly is only rarely found in other tumors [3].

In this study we were impressed by the large irregular clumps of calcific material which we believe represent necrotic bone sequestrated and displaced by the soft issue mass. Finer areas of dystrophic calcification tend to be found in smaller, less advanced tumors and were relatively uncommon in this study.

Rarely, bone destruction may not be present or not be recognized. In one patient in this study with a 12 cm soft tissue mass, high resolution CT and magnetic resonance imaging failed to demonstrate convincing evidence of bone destruction. This unusual tumor, regarded as an anaplastic chordoma, showed a 2 cm area of bone destruction at pathological examination. Because of the many unusual features of this tumor it is being reported elsewhere.

In case 3, following resection of the primary tumor which showed bone destruction, the extensive recurrence occurred purely in the soft tissues of the pelvis and abdomen. The residual sacrum and lumbar spine showed no evidence of bony involvement. The bony margins were clear at the original surgery but soft tissue tumor must have remained. This emphasizes the importance of clearing the pelvis of all soft tissue tumor at the time of surgery. Occasionally, however, a chordoma without any bony destruction even at pathological examination may be found. Willis [28] has described such a case.

The behavior of the tumor is unpredictable and differs in this respect from other malignant tumors of bone. Some large tumors which appear aggressive may lead to survival for a decade or more. Others which appear relatively small and innocuous may result in death within a few years. Case 3 is a case in point. Patients usually die from local disease but metastases are not uncommon. For many years metastases to distant sites were thought to be distinctly uncommon and the incidence to be below 10%. In this study the figure was 24% and recent studies have confirmed this figure [5, 27]. Metastases may occur at any time during the course of the disease and are commonly asymptomatic. In one of the patients in this study who died 20 years following original diagnosis, pulmonary metastases (unsuspected) were found at autopsy. However, since few patients appear to die from their metastases it is fair to question the significance of metastatic disease in this tumor.

The experience with magnetic resonance imaging (MRI) in sacral chordoma is limited. A recent report [19] described four such patients. The advantages at this time over CT are not great and two patients who were thought to have rectal in-

volvement by the tumor on CT showed similar suspicious findings on MRI. In neither case was there rectal invasion at surgical exploration. However, the direct imaging in coronal and sagittal planes do offer considerable advantages, and with technical advances in imaging with MRI this is likely to become the modality of choice in the future. High resolution CT does provide accurate information regarding the spread of chordoma in the bone and soft tissues, and comments by Hudson and Galceran [11] regarding the unreliability of CT in evaluating soft tissue spread are probably related to early generation CT images in their study.

The management of patients with large sacrococcygeal chordomas is frequently difficult. The optimal form of treatment is wide surgical resection leaving tumor-free margins. Radical resection is, however, frequently not possible because of the massive size of the tumor. In 12% of the patients in this study surgery was not even attempted. Of those that underwent surgery – usually palliative – 80% of tumors recurred at least once. Over the past decades a variety of surgical approaches have been described [12, 15].

At this center the standard approach has been a preliminary celiotomy with ligation of the internal iliac and median sacral vessels. There is good visualization of the soft tissue mass and good exposure of the pelvic viscera, and the rectum and bladder can be completely cleared of soft tissue tumor [2]. The sacrum and the associated tumor is then resected from a posterior incision. For many years it was regarded as imprudent to resect chordomas above S2 because of probable damage to the sacral nerves and denervation of the bladder and bowel. Also instability was feared.

Stener and Gunterberg [23, 24] have recently described a far more radical operation in which high sacral resections can be achieved without loss of pelvic stability. Control of bowel and bladder, and the management of impotence can be achieved with a variety of devices. These complications, however, may make this type of surgery unacceptable to many patients. Preliminary results in a relatively small number of patients are encouraging but further follow-up is necessary for evaluation of this radical surgery.

Radiation therapy has long been part of the routine treatment of sacrococcygeal chordoma. Small tumors involving the distal sacrum and coccyx can probably be cured by irradiation alone but they are more satisfactorily dealt with by surgery. In bulky tumors there is little objective evidence of a decrease in tumor size following radiation. However, good subjective responses do occur. Beneficial responses are more likely to follow treatment doses above 6,000–8,000 cGy [16] and Saxton [20] advocates radiation doses from 7,000–8,000 cGy. Irradiation is almost always given following surgical excision of the tumor, whether radical or palliative. In a recent study, irradiation prior to surgery in a small group of patients produced no significant change in radiation response [18]. Recurrences are common in this tumor and many courses of irradiation have been given over long periods with good palliation. Soft tissue radiation necrosis in the pelvis has been described by several authors and is not without risk. A rare complication of radiation therapy is radiation-induced sarcoma. This occurred in case 2 in this study, and a recent case of malignant fibrous histiocytoma has been reported [6].

In a recent study [1], there was no difference in survival behavior of the patients who had palliative radiation and those who were not treated. There was, however, a significant difference in survival between the group who received radical surgery and radiation therapy and the group who received palliative surgery and palliative radiation therapy. A variety of chemotherapeutic drugs, either singly or in combination, have been tried in this tumor. There is little evidence that they have been effective and no studies of more than occasional cases are available. Reports in the literature are largely anecdotal [22].

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