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Two-stage Therapy in the Treatment of Sacral Tumors

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Summary. Sacral tumors are rare and may be clinically overlooked for a long period, because the symptoms and signs are often mild and non-specific. This led to frequent errors in clinical diagnosis and a long delay between the onset of symptoms and treatment. On presentation the lesions frequently expanded the anterior cortex, however, in most patients the periosteum of the sacrum and the presacral fascia form an unbroken barrier for a tumor. The pelvic viscera are not infiltrated by the tumor until late. Wide excision is difficult and often causes urogenital and/or anorectal dysfunction, but preserving the sacral nerve roots often leads to local recurrence. Surgical wide excision with a combined anterior-posterior approach is considered the treatment of choice for large lesions with significant anterior intrapelvic extension. This paper reports data resulting from the treatment of five large sacral tumors with comments on the results.

Zusammenfassung. Sakraltumoren sind selten und können klinisch über einen langen Zeitraum hinweg übersehen werden, da die Symptomatik oft schwach ausgeprägt und unspezifisch ist. Dies führte klinisch häufig zu Fehldiagnosen und zu einer langen Verzögerung des Therapiebeginns seit dem Einsetzen der Symptome. Bei der Darstellung der Läsionen zeigt sich häufig, daß diese die vordere kortikale Wand ausdehnen oder perforieren. Bei den meisten Patienten bilden das Periost des Sakrums und die präsakrale Faszie eine nicht zu durchbrechende Barriere für einen Tumor. Die Beckeneingeweide werden erst spät von dem Tumor infiltriert. Eine weite Resektion ist schwierig; es kann dabei zu einer Blasen- und/oder Darmfunktionsstörung kommen. Erhält man aber die sakralen Nervenwurzeln, führt dies oft zu lokalen Rezidiven. Bei großen Läsionen mit weiter Ausdehnung ins kleine Becken wird die radikale Resektion mit einem kombinierten antero-posterioren Zugang als Therapie der Wahl angesehen. In der vorliegenden Arbeit wird über die Behandlungsergebnisse von fünf großen Sakraltumoren berichtet und kommentiert.

Large sacral tumors are uncommon and may require a considered surgical anterior-posterior approach; but this decision is influenced by the type, the location and expansion of the lesion as well as the potential loss of adequate urinary and rectal continence. The main problems are associated with locally aggressive tumors. Benign sacral lesions are in most cases giant cell tumors, aneurysmal bone cysts or osteoblastoma and malignant lesions are either chondrosarcoma or chordoma [4, 17], which are located adjacent to important pelvic viscera and which, if not exstirpated, cause local damage and dysfunction of visceral and/or bony structures. Radical surgical excision is the primary treatment of choice for these tumors, while susceptability for chemotherapy and radiotherapy is low [8, 12, 16, 17] and radiation-induced sarcoma have been reported [2, 3]. Although several surgical and non-surgical techniques have been advocated [9–11, 13–16], there are no extensive experiences or clinical trials to evaluate the effects of these modalities. Stener and Gunterberg [15] reported a two-stage (anterior-posterior) procedure for exstirpation of sacral tumors. The method generally consists of high radical sacretomy either above or through the foramina of the first or second sacral nerve root. They believe that only radical excision in which a complete resection of

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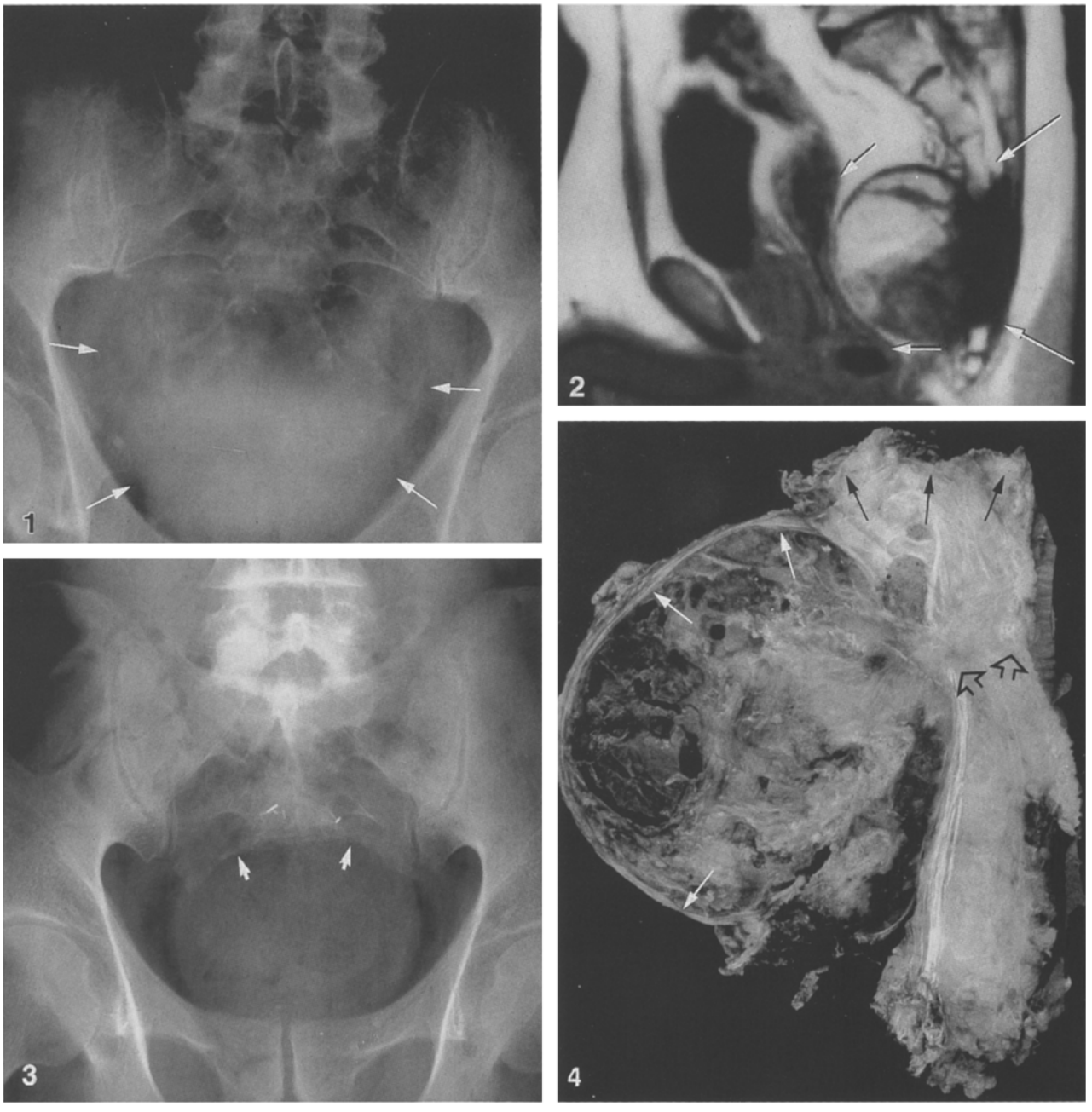


Figs. 1–4. Case 4. **Fig. 1.** Preoperative X-ray showing the destruction and anterior extension (white arrows) of the chordoma

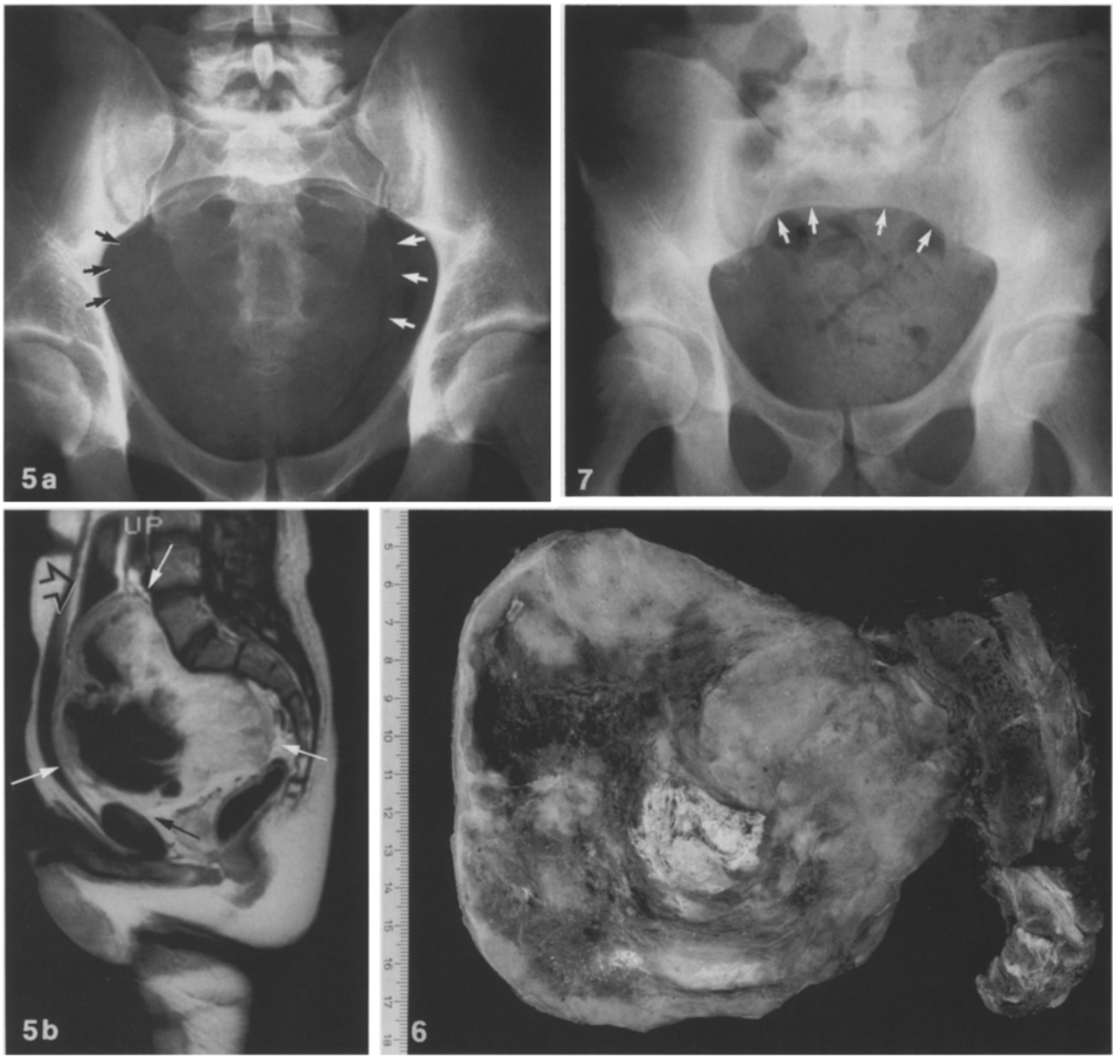
Fig. 2. NMR-scan showing large anterior mass with destruction of the S3 to S5 sacral segments extending posteriorly into the soft tissues (big arrow). Note the rectum is displaces anteriorly (small arrows)

Fig. 3. Roentgenograph 4 months postoperatively. The white arrows indicate the superior margin at the S2/S3 level

Fig. 4. Median sagittal section of the specimen showing the proximal margin (black arrows) and the intact presacral fascia (white arrows). The tumor destructs the anterior and posterior sacral cortex (open arrows)

the tumor is achieved, yields long term disease-free intervals.

The following presentation is based on clinical material from 5 patients. In one of the five patients (case 1, Table 1) local recurrence after a posterior approach was the indication for a two stage procedure; the other four patients (cases 2–5, Table 1) were initially treated with the combined anterior-posterior technique. Two sacral resections were carried out for chordoma (case 1, 4), one for a giant cell tumor (case 3), one for a chondrosarcoma (case 2), and one for a low-malignant schwannoma (case 5). All patients are well and free of tumors or metastases.



Figs. 5–7. Case 5. **Fig. 5. a** Anterior-posterior roentgenograph of patient in case 5 showing the anterior extraskeletal part of the tumor. Note the intact sacrum and calcification around the periphery. **b** NMR-scan showing a huge tumor occupying almost the whole lower pelvic (thin arrows) reaching the pubic bone and abdominal wall anteriorly and the L4/L5 level superiorly (thin arrows). Note the displacement of the bladder (open arrows)

Fig. 6. Median sagittal section of the specimen showing the base and the anterior part of the lesion. Note the almost intact sacrum

Fig. 7. Roentgenograph 3 months postoperatively. The arrows indicate the level of amputation between S1/S2

Case Reports

Case 4

A 56-year-old man complained of gluteal pain for two years when abdomino-rectal palpation revealed a large intrapelvic tumor. CT, NMR and roentgenographic examination showed a sacrococcygeal tumor destroying the S3–S5 vertebra (Fig. 1, 2). Open dorsal biopsy revealed the histologic diagnosis of a chordoma. An amputation between S2/S3 bilaterally, leaving the S1 and S2 nerves intact, was performed. Four months later the gentamycine beans were removed (Fig. 3). Macro- and microscopical inspection of the $17 \times 15 \times 10$ cm specimen proved to be radically (Fig. 4). At follow up two years after surgery, the patient is well, continent as to bowel and bladder and returend to his previous job.

Case 5

A 25-year-old man consulted several doctors for 2½ years, complaining of vague intrapelvic pain and of micturation disturbances for several weeks. Physical examination revealed a suprapubic tumor, bulging the anterior abdominal wall, reaching the L4/L5 level. No neurological deficit could be established. CT, NMR and roentgenographic examinations revealed a huge tumor reaching the pubic bone anteriorly and the L4/L5 level superiorly (Fig. 5). The bladder was displaced superiorly and the rectum latero-posteriorly. A chordoma was suspected and a two-stage sacral amputation through the S1/S2-level, preserving the left S 1 nerve, was planned and performed. Histological examination showed a low malignant schwannoma developed from the second right sacral nerve. The margins were free of tumor tissue (Fig. 6). Five weeks postoperatively the gentamycine beans were removed (Fig. 7). The patient has a permanent bowel and bladder dysfunction, but is continent. At follow-up sixteen months after the operation the patient is well and in revalidation.

Results

Among the 5 patients, 1 patient (case 1) had a previous subtotal excision and additional radiotherapy (60 Gy) for a chordoma. He presented with a recurrent tumor 1½ years after the first surgical procedure (Table 1).

On first presentation, all patients had no objective neurological deficit by physical examination. Four patients (cases 1–4) complained of low back pain or gluteal pain, one of them had radiating pain to the left leg (case 3). Three patients (cases 1, 2 and 5) had neurological bladder dysfunction as judged by cystometry. All patients had extensive lesions (ranging from 13 to 26 cm) which protruded into the presacral soft tissues, only well delimited by a presacral fascia. In cases 1–4 the lesion involved the entire vertebra (body and posterior elements) and infiltrated the dorsal structures too. In case 5 the tumor produced only an anterior enlargement and extended from the S2 anterior right foramen up to L4/L5. The definite surgical treatment consisted of the two-stage wide excision of the tumor through S1 (case 3), between S1/S2 (case 1, 2, 5) and between S2/S3 (case 4). In case 1 to 3, the first sacral nerves were bilaterally preserved. In case 5 only the left S1 nerve could be preserved and in case 4 both S1 and S2 nerves. In the postoperative period all patients had bowel and bladder dysfunction therapied with diet and temporary urinary catheterisation.

Intraoperatively, there were no complications, in the postoperative period two hematoma, one bowel hernia, two urether dysfunctions and one peroneal paralysis was reported. No wound infection, especially none after closure of the posterior wound, was reported.

At follow-up all patients had on indication standard radiographs, CT scans or bone scans. Until now no local recurrence or metastases could be diagnosed, all patients are pain-free and returned to their preoperative daily activities.

Discussion

Wide curative surgical resection is precluded in many patients with sacral tumors in respect to the localisation and extension of the tumor, for fear of sacrificing normal urogenital and anorectal function and the impairment of the pelvic girdle stability. Some authors advocate radiotherapy for both malignant and aggressive benign lesions and for palliation of recurrent tumors [4, 6, 12, 13, 16]. However, these tumors are almost radioresistant and radiation has a significant morbidity with regard to the tolerance of the viscera and the spinal cord. Other treatment modalities include the use of cryosurgery and several different wide local excision techniques. Cryosurgery can have a place in the treatment of sacral tumors either curative or palliative in situations where a resection might result in bony instability [1, 4, 18]. Although the sacrum need not to be resected, serious problems such as skin and tissue necrosis, susceptability of cryolised tissue for infection, and loss of control of the extend of the cryolesion can complicate the treatment [18]. A large lesion (e.g. chordoma >10 cm) can only be treated palliatively [18].

McCarthy et al. [11] described a strictly posterior approach, Hays [7], Cody et al. [1] and Localio et al. [10] described different combined abdominal-sacral approaches for the treatment of sacral tumors. Detailed surgical techniques for high radical sacral amputation was reported from Stener and Gunterberg [15].

For a detailed description of these techniques we refer to the authors' original paper [15]. Essentially the first stage of the anterior-posterior procedure is the ligation and resection of the internal, lateral and median sacral arteries and veins followed by an osteotomy of the anterior cortical wall of the sacrum at the desired level. As we have seen an abdominal hernia with impairment in voluntary voiding at the site of the dissection of the rectus abdominus tendons (case 2), we have modified the incision for the anterior approach (Fig. 8). We prefer two separate incisions through the skin, the subcutaneous layers and along the lateral border of the aponeurotic sheath of the rectus abdominus. The peritoneal cavity need not to be opened. Even in very large lesions (case 4, 5) a good exposure of the intrapelvic structures is obtained and the rectus abdominus muscles keep strong and functional. As for stool and urine continence the

Table 1. Clinical data on patients with sacral tumors

Patients (age/sex)	Symptoms	Level	Size (cm)	Therapy	Postoperative status	Complications	Histological diagnosis	Follow-up
1. 64/♂	Pain in gluteal region, micturation disturbances	Recurrent tumor at S2 level, 1.5 yr after dorsal amputation between S3/S4 + radiotherapy (60 Gy)	15	Amputation between S1/S2 bilaterally	Permanent bowel and bladder dysfunction, not continent Uses a cane	Pneumonia	Recurrent chordoma	2.5 yr disease free, no metastases
2. 25/♂	Lower back pain, progressive bowel and micturation disturbance	S2/S3	14	Laminectomy S2/S3 (neurosurgeon) Amputation below S1/S2 bilaterally + radiotherapy (56 Gy)	Permanent bowel and bladder dysfunction,	Abdominal hernia	Chondrosarcoma	4.0 yr disease free, no metastases
3. 24/♀	Pain in gluteal region, radiating to the leg	S1	13	Amputation through S1 + Palacos	Permanent bowel and bladder dysfunction, continent Pelvic girdle instability Uses a cane and pelvic bandage	Left peroneal pareses Temporary left ureter dysfunction	Giant cell tumor	3.5 yr disease free, no metastases
4. 50/♂	Gluteal pain	S3	17	Amputation between S2/S3 bilaterally	Temporary bowel and bladder dysfunction, continent	Ventral hematoma	Chordoma	2.0 yr disease free, no metastases
5. 25/♂	Micturation disturbance	S2	26	Amputation between S1/S2 bilaterally	Permanent bowel and bladder dysfunction, continent	Ventral hematoma Temporary right ureter dysfunction	Low malignant Schwannoma	1.6 yr disease free, no metastases

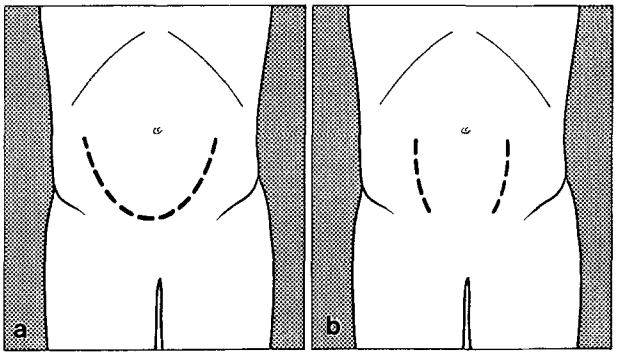


Fig. 8a, b. Demonstration of the ventral-abdominal skin incision according to Stener (**a**) and the modified technique (**b**)

patients need and must use abdominal muscles and diaperse to empty the bladder and bowel regularly. The second dorsal procedure can be done several days or weeks later. A posterior midline incision with division of muscles and ligaments is performed, the dural sac exposed, ligated and devided at the desired level and the osteotomy of the sacrum is completed. Once all sacral, pelvic and visceral structures have been ligated and divided or dissected and retracted the specimen can be removed. The closure of the posterior wound can be a serious problem. After removal of the specimen a large cavity remains normally and must be drained for a considerable period of time. Infection and delayed wound healing often complicate the postoperative course [15, 18]. We therefore filled the cavity with a bag of gentamycine beans and closed the wound primarily. This method appeared to be safe and obtained a primary wound healing in two to three weeks in all patients. The gentamycine beans were removed one to several months later. At operation the cavity was almost complete obliterated and filled with granulation tissue.

High sacral amputation through the first sacral vertebra weakens the pelvic girdle stability by approximately 50%; a vertical load without breaking is resisted [5, 15]. Only one patient complained of pelvic girdle instability after high sacral amputation (case 3). A significant improvement could be registered (EMG, walking analysis) when wearing a special pelvic girdle orthesis.

High sacral amputation may cause significant neurological deficits. Preservation of the first two sacral nerve roots (S1, S2) bilaterally retains the bladder and rectal function in practically all cases. The additional bilateral sacrifice of the S2 nerves gives a complete bladder, urethral and rectal sphincter denervation [4, 14, 15]. Its effects on the motoric and sensoric function of the lower extremities is minimal. Walking is almost normal; only the gluteal muscles are partially denervated, but can be compensated. All patients have a permanent or temporary bowel and bladder dysfunction postoperatively, only one patient (case 1) is not continent. All other patients have regained fecal and urinal continence by diet and regular voiding.

Although conclusions are premature in respect to the patient collective and follow-up, it is our opinion that the treatment of choice for these primary and recurrent sacral tumors should be a primarily radical surgical excision. Radiation therapy is indicated for inoperable cases only. The success of a surgical treatment lays in a carefully planned procedure (CT, NMR, arteriography), the experiences of the surgeon and the information and instruction of the patient.

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