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Presacral tumors. Analysis of 20 surgically treated patients

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A B S T R A C T

Introduction: Presacral tumors are a rare and little known pathology. We present our experience in its surgical treatment.
Methods: Analysis of a series (1995–2008).
Results: Twenty patients (14 women and 6 men), average age 46 (29–71) years. Patients with benign tumors were younger (43.5 [10.2]) years that those who had malignant tumors (62 [10.7]; P=.002). The median duration of symptoms was 1 year (2 month–50 years). The first symptom was suppuration, palpable mass or constipation in 10 cases or constipation, abdominal pain in 4, back pain in 4 and a casual finding in 3. The surgical approach was perineal in 9 (45%), abdominal in 8 (40%), and combined in 3 (15%) cases. Fifteen (75%) tumors were benign, with teratomas being the most frequent (5 cases). Another 5 were malignant: 2 chordomas, 1 malignant cystic teratoma, 1 malignant mucinous cystadenocarcinoma, and 1 lymphoma. Mean postoperative stay was 6.6 (5) days. There were post-surgical complications in 5 (20%) patients. After an average follow-up of 3.5 (4) years, 2 patients died (malignant teratoma and cystadenocarcinoma, respectively) due to morbidity and to a non-resectable recurrence, and a benign schwannoma has recurred, the rest being free of disease.
Conclusions: Surgical treatment by a group of surgeons experienced in perineal and pelvic surgery can obtain good results with few complications, with the exception of some malignant tumors, depending on the infiltration of adjacent structures and their histological type.

Tumores presacros. Análisis de nuestra experiencia en 20 casos tratados quirúrgicamente

Palabras clave:
Tumores presacros
Tumores retrorrectales

R E S U M E N

Introducción: Los tumores presacros son una enfermedad infrecuente y poco conocida. Presentamos nuestra experiencia en su tratamiento quirúrgico.
Métodos: Análisis de una serie (1995–2008).

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Teratoma
Cordoma
Abordaje parasacro

Resultados: Estudiamos a 20 pacientes (14 mujeres y 6 varones), media de edad de 46 (29–71) años. Los pacientes con tumores benignos tenían una media ± desviación estándar de edad (43,5 ± 10,2 años) menor que los que presentaban tumores malignos (62 ± 10,7; p = 0,002). La mediana de duración de los síntomas fue de 1 año (2 meses–50 años). El primer síntoma fue supuración, masa palpable o estreñimiento en 10 casos; dolor abdominal en 4, lumbalgia en 4 y hallazgo casual en 3. El abordaje quirúrgico fue perineal en 9 (45%) casos, abdominal en 8 (40%) y combinado en 3 (15%). Hubo 15 (75%) tumores benignos; los más frecuentes, los teratomas, con 5 casos. Otros 5 fueron malignos: 2 cordomas, 1 teratoma quístico maligno, 1 cistoadenocarcinoma mucinoso y 1 linfoma. La media de estancia hospitalaria fue 6,6 ± 5 días. Hubo complicaciones posquirúrgicas en 5 (20%) casos. Tras una media de seguimiento de 3,5 ± 4 años, fallecieron 2 pacientes (teratoma maligno y cistoadenocarcinoma) por comorbilidad y recidiva inextirpable, respectivamente, y ha recidivado uno benigno (schwannoma); los demás están libres de enfermedad.

Conclusiones: El tratamiento quirúrgico realizado por un grupo de cirujanos experimentado en cirugía pelviana-perineal puede obtener buenos resultados con pocas complicaciones, a excepción de algunos tumores malignos, según la infiltración de estructuras vecinas y su tipo histológico.

Introduction

Retrorectal tumours are highly infrequent occurrences, usually benign and located in the area known as the presacral or retrorectal space. The tumours may derive from remains of embryological structures or may be a consequence of malignization in the area.[1] In general, they are asymptomatic or have unspecific symptomatology, making their diagnosis difficult and on occasion by chance. The fact that the area where they reside is asymptomatically occult and difficult to access makes experience in surgical intervention a requisite for treating them. A good knowledge of its peculiarities is also important. This article discusses an analysis from our experience of their surgical treatment with 20 operated cases.

Material and methods

We carried out an observational study, with prospective collection of data, of a group of 20 patients with tumours in the retrorectal space. They were resected by the same surgical team in the Hospital of Sagunto and General University Hospital Consortium of Valencia from 1995–2008. Variables studied were: symptoms, diagnostic exams, surgical treatment, and immediate and later results. Data obtained were analyzed by the SPSS® version 13 for Windows statistical package (SPSS Inc., Chicago, United States). Quantitative variables were analyzed by the student *t* test, when they were adjusted to a normal distribution, and the Mann-Whitney test, in the contrary case. A *P* value less than .05 was considered statistically significant.

Results

We studied a total of 20 patients (6 men and 15 women), with a median age (IQR) of 46 (37–53) years. A total of 7 were treated in the Hospital of Sagunto, from 1995 to 2005, and the other 13, in the General University Hospital Consortium of Valencia, from 2005 to 2008, by the same surgical team; 11 (55%) of patients were sent from other centres to our coloproctology unit for treatment.

Fifteen patients presented with benign tumours and 5 with malignant ones; the average (standard deviation) of age was significantly lower in patients with benign tumours (43.5 [10.2] vs 62 [10.7] years) (*P*=.002). Only 1 (7%) of the 15 women presented with a malignant tumour versus 4 (80%) of the 5 men (*P*=.004). Median evolution of symptoms was 1 year (interval, 2–50 years). The benign tumours were diagnosed with less time of evolution than the malignant ones, an average of 5 (10) versus 12 (21) years after the beginning of symptomatology, and this difference was not significant (*P*=.266).

The most frequent initial symptom was a perineal mass in 5 (25%) cases. In 3 patients the mass was related to perineal pain, and surgical drainage was carried out because the tumour was mistaken for a perianal abscess (2.3 drainages on average). Later the patients presented with a maintained suppuration. Other symptoms were low backache in 4 (20%) cases, abdominal pain in 3 (15%) and perineal pain in another 3, evacuation difficulty in 1, and spontaneous rectal suppuration in 1. Three (15%) patients were diagnosed by chance when being examined for other reasons. One patient presented with a reducible perineal tumour which corresponded to a retrorectal tumour herniated because of the musculature of the pelvic floor.

Prior to this, 4 patients had been operated on in other centres: two teratomas, a cystadenocarcinoma, and a schwannoma. Resections were carried out on 3, with healing criterion in 2 teratomas and palliative criterion in the cystadenocarcinoma, with an average of 1.6 interventions. Schwannoma was considered inoperable, and a simple biopsy was carried out.

All tumours were substantial in the digital rectal examination, except for one of the chordomas. Preoperative evaluation consisted of a computerized tomography (CT), magnetic resonance (MR), or both in all cases (Figures 1

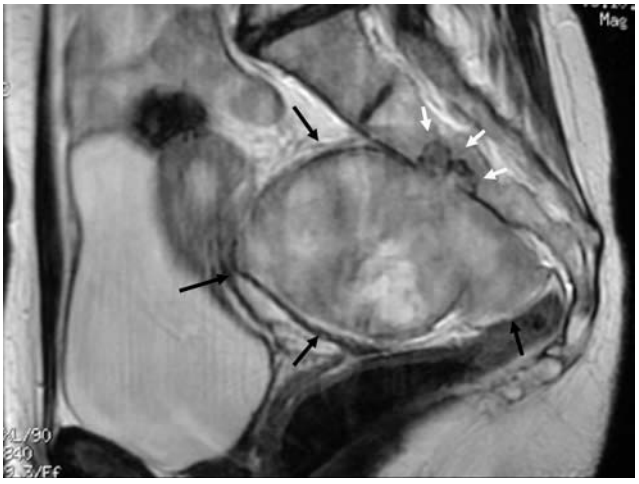

Figure 1 – Magnetic resonance image of a schwannoma (black arrows), with sacral involvement (white arrows).

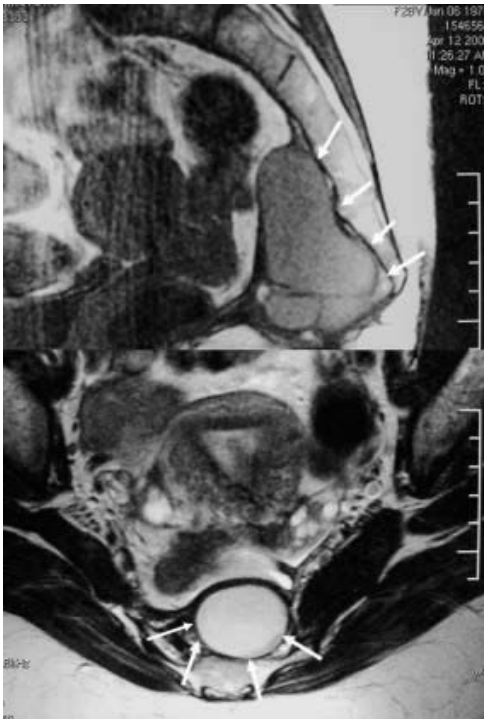

Figure 2 – Magnetic resonance image of a recurrent cystic teratoma (arrows).

and 2), and an endoscopic study, at least a rectosigmoid also in all except for 1 patient, and an endorectal ultrasound in 7 cases.

Criterion for selecting the surgical route was the upper edge of the tumour, and thus, the tumours found over S2-S3, or those with the upper edge not being detected in the digital rectal examination were operated by the abdominal route, and in the contrary case, perineal route. A mixed approach was carried out for large tumours and chordomas. Consequently, 9 (45%) were operated by perineal route; 8 (40%), abdominal; and 3 (15%) cases (2 chordomas and a gigantic teratocarcinoma) by combined route. A tumour resection with expected healing capacity was carried out on 17 (85%) patients. Only 1 intraoperative biopsy was taken in a lymphoma, and in 2 patients, the surgery was palliative: a relapsed cystadenocarcinoma, which fistulized the rectum and broadly infiltrated the gluteus musculature and pelvic vasculonervous structures, and a large-sized teratocarcinoma.

In all cases, the abdominal procedure was a median laparotomy, and the perineal procedure, York-Mason parasacral access with sphincter preservation (Figure 3). Two chordomas required a partial sacrectomy and the combination of a colostomy and plastic reconstruction of the perineum. The average postoperative stay was 6.6 (5) days, with an interval of 2–25. Patients with malignant tumours had a longer stay than those with benign tumours, although without reaching statistical significance (11.4 [8] vs 5.1 [2] days; *P*=.15). Postoperative morbidity consisted of 5 (25%) complications: two seromas from a parasacral lesion, 1 mesh infection which required reintervention, 1 partial dehiscence of the detached tissue of a chordoma, and 1 persistent pain in the lower extremity after excision of a large pelvic schwannoma, which disappeared after 1 year.

Seventy-five percent of tumours were benign; the most frequent of them was the teratoma, with 5 cases; 5 tumours were malignant, 2 of them were chordomas (Table 1).

After an average follow-up of 3.5 (4) years (2 months to 13 years), the patient with teratocarcinoma, subject to a palliative resection, died due to unrelated causes to the illness. The patient with mucinous cystadenocarcinoma, also with palliative resection, died at ten years of diagnosis due to a massive pelvic infiltration after multiple interventions for reducing the tumour mass. One benign tumour recurrence (7% of benign patients) occurred; 1 sacral schwannoma occurred which will soon be operated on (Table 2). There were no postoperative alterations in faecal continence and none in evacuation habits of operated patients with benign tumours, and constipation improved in the 4 who presented with this in the preoperative. Regarding malignant tumours, a neuromodulation of sacral roots was carried on a patient with sacral chordoma 3 years after the intervention, for whom they respected an S3 root. Later, the colostomy was closed, and the patient currently presents with mild faecal incontinence.

Discussion

The presacral or retrorectal virtual space is limited from behind by the presacral fascia and in the front by the fascia propria of the rectum. Reflection of the pelvic peritoneum is its upper limit, and the Waldeyer fascia, the lower, which separates it from the supralevator space. Finally, the ureters, iliac vessels and sacral nerve roots constitute its lateral limits. Its content includes tissues of heterogenous histological origin, because during the first stages of embryological development, it is occupied by pluripotential cells which differentiate toward different histological classification (vascular, nerve,

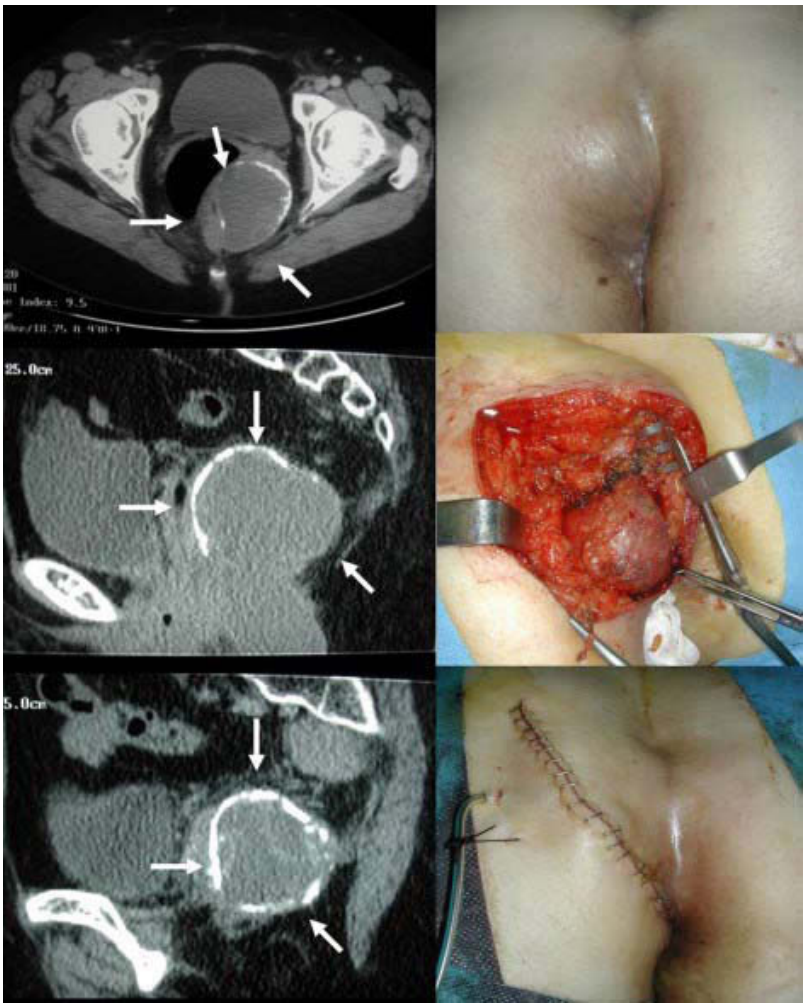

Figure 3 – Computerized tomography images and magnetic resonance of a teratoma with calcified areas (left). Physical examination and parasacral procedure (right).

Table 1 – Pathological anatomy

	No. (%)
Benign	15 (75)
Teratoma	5 (25)
Schwannoma	3 (15)
Hydatid cyst	2 (10)
Cystic lymphangioma	1 (5)
Epidermoid cyst	1 (5)
Angiolipoma	1 (5)
Angioleiomyoma	1 (5)
Fibrous tumour	1 (5)
Malignant	5 (25)
Chordoma	2 (10)
Teratocarcinoma	1 (5)
Cystadenocarcinoma	1 (5)
Lymphoma	1 (5)

connective, osseous, etc), and therefore, they may condition the appearance of an extensive range of tumours.[2]

These are infrequent lesions, with an incidence around 0.02%, and less than 1 in every 40 000 hospital admissions.[1,3] Therefore, studies from the literature are almost always retrospective and with a small sampling size, such as this sample, consisting of 20 patients who were intervened over 14 years, and this is one of the largest described in the literature.

These conditions may be classified by separating between congenital or acquired, and benign or malignant conditions. Furthermore, they may be divided into solid lesions, such as the teratomas, chordomas, schwannomas or ependymomas, or cystic lesions, such as *tailgut cysts* or dermoid cysts (Table 3). The most frequent tumours are congenital and represent 2/3 of all cases, and benign tumours make up 66% of these.[3] Seventy-five percent of the excised neoplasias in our study were benign, with teratomas being the most common tumours. Chordoma was the most frequent malignant tumour in our study.

Presacral tumours were more common in women.[1-4] However, of those, the most frequent among malignant tumours were chordomas generally found in the men. Age distribution was broad in our case, and ranged from 29 to 71 years, and there is no predominant age in symptom presentation. Diagnosis may be made from the prenatal

Table 2 – Characteristics of patients

No.	Age	Gender	Year of surgery	Time of evolution, y	First symptom	CT	MR	Procedure	Pathological anatomy	Recurrence	Follow-up, y
1	40	W	1995	3	Abdominal pain	Yes	No	A	Cystic lymphangioma	No	13
2	34	W	1996	30	Perineal pain	Yes	No	P	Teratoma	No	12
3	52	W	1998	1	No	Yes	No	P	Teratoma	No	10
4	63	W	2000	0.2	Abdominal pain	Yes	No	A	Hydatid cyst	No	8
5	70	M	2004	7	Suppuration	Yes	Yes	P	Mucinous cystadenocarcinoma	Yes	4
6	71	M	2004	50	Weight	Yes	Yes	A-P	Malignant teratoma	Yes	2
7	37	W	2005	0.2	No	No	Yes	A	Schwannoma	Yes	3
8	46	W	2005	5	Perineal pain	Yes	Yes	P	Epidermoid cyst	No	3
9	56	W	2006	2	Weight	Yes	Yes	P	Cystic teratoma	No	2
10	38	M	2006	0.2	Abdominal pain	Yes	No	A	Hydatid cyst	No	2
11	68	W	2006	2	Low backache	No	Yes	A-P	Chordoma	No	2
12	53	M	2005	0.3	Low backache	Yes	Yes	A-P	Chordoma	No	3
13	46	W	2006	1.5	Perineal pain	No	Yes	P	Angiolipoma	No	2
14	56	M	2007	0.2	Low backache	No	Yes	A	Schwannoma	No	1
15	29	W	2007	29 (2 recurrences)	Weight	No	Yes	P	Cystic teratoma	No	1.6
16	36	W	2007	1	Reducible mass	No	Yes	A	Angioleiomyoma	No	1
17	29	W	2008	0.3	Constipation	Yes	Yes	P	Cystic teratoma	No	0.5
18	52	W	2008	0.5	Weight	Yes	Yes	P	Fibrous tumour	No	0.3
19	42	W	2008	0.8	No	Yes	Yes	A	Schwannoma	No	0.2
20	48	M	2008	1	Low backache	Yes	Yes	A	Lymphoma	No	0.2

A indicates abdominal; A-P, abdominal-perineal; M, men; P, perineal (parasacral); W, women.

Table 3 – Types of presacral tumours

Congenital	*Inflammatory*
Developing cysts	Granulomas
Epidermoid cyst	Abscesses and fistulas
Dermoid cyst	Osseous
Cystic hamartoma (tailgut)	Osteoma
Teratoma	Osteogenic sarcoma
Chordoma	Ewing tumour
Anterior meningocele	Gigantic cell tumour
Rectal duplication	Chondromyxosarcoma
Tumours of adrenal remains	
	Miscellaneous
Neurogenic	Metastasis
Neurofibroma	Lymphangioma
Neurilemmoma	Desmoid tumour
Ependymoma	Leiomyoma
Ganglioneuroma	Fibrosarcoma
Neurofibrosarcoma	Lymphoma
Schwannoma	Hydatid cyst

period to very advanced ages.[1,4,5] In any case, in our study, older age and male gender show a significant relationship with histological malignity.

Classically, diagnosis is late due to a lack of specificity or absence of symptoms.[1,2,4] It is certain that clinical presentation of these neoplasias leads to differing diagnoses of the most frequent diseases, mainly with fistulas, perianal abscesses or pilonidal cysts.[6]

Symptoms tend to be related to location, size, and presence or absence of infection in the case of cystic tumours, which are described in 30% of cases.[7] In our study, 3 patients presented with an infection which required surgical drainage in 2 of these cases, and in another case, the rectum drained spontaneously, which means 20% of infected cystic tumours. Pain is the most common symptom and generally indicates a bad prognosis, as it is twice as frequent in malignant neoplasias than in benign ones.[1,8,9] Patients may complain of a sensation of a mass or perineal or lumbar heaviness, which typically worsens in the seated position.[3,8] Of those included in our study, the majority (85%) presented with some symptom at the time of diagnosis, but this was highly unspecific, ie, lumbar pain, abdominal irritations or the sensation of a perineal mass, as documented by other authors.[1,4,10] Other symptoms such as constipation, urinary or faecal incontinence, alterations in urination or dystocia, tend to be typical of advanced tumours which produce compression of neighbouring structures.

The digital rectal examination is mandatory, because the majority of these lesions are accessible by this examination. In the largest study published, that of the Mayo Clinic, 97% of patients presented with palpable tumours in the digital rectal examination; this is confirmed by other authors.[1,4,11] Our study had a 95% rate, as only one of the tumours was not detected by this examination.

The additional tests are very useful. A simple x-ray of the pelvis may show bone expansion and destruction, typical of malignant lesions, but it may also show calcifications and tumours which occupy soft parts.[8] However, to find precise information relating to localization and relationship with the rest of the pelvic structures, we use on imaging techniques, which are now tests of choice. Consequently, the endorectal ultrasound is useful for characterizing the quality of presacral tumours, such as their relationship with the layers of the rectum.[9] In addition, there has been a drastic change in diagnostic approach, since tests with greater benefit, such as the CT or MR are chosen directly. These techniques allow for differentiation between solid, cystic or mixed tumours, along with the unique advantages of each one. Diagnostic accuracy but also the choice of surgical approach may depend on these techniques, and for example, the MR shows soft parts with greater resolution, which may be essential for tumours of neural origin, implying an alteration of the thecal sac, or medullary or nerve compressions.[3,6,10,11] At least one or another was used in all cases in our study.

One subject for debate is the role of preoperative biopsy, because it runs the risk of producing tumour spreading due to the puncture route in the case of a solid tumour or its infection, which is lethal on occasion if we are faced with liquid characteristics and if it corresponds with a meningocele.[3] The majority of the time, the approach when facing this problem does not change, and furthermore, rupture of the tumour capsule may compromise the possibility of the patient's recovery.[8] Because of this, we, along with other authors, do not believe this to be recommendable, except for inoperable tumours,[4,12] in which case, an appropriate adjuvant treatment could be adapted. In the case of carrying this out, transrectal, transvaginal, or transperitoneal procedures should be avoided, and the puncture route should be included as a resection sample. The transperineal and/or parasacral routes are more effective, less damaging and have less risk of infection for obtaining the puncture route sample.[3,6,8] In our study, a preoperative biopsy was carried out on 2 patients in other centres before being sent to us.

Once a retrorectal tumour has been diagnosed, it should be excised,[1,4,8,13] because the lesion may be malignant at the time of diagnosis or because of the risk of degeneration, especially teratomas in pediatric patients.[14] In young women, presacral masses may cause dystocia. Cystic lesions have an occasional risk of very serious infection, and this may make complete excision difficult, decrease possibilities of recovery and increase the rate of recurrences.[1,15,16]

The surgical approach of these tumours may depend on various factors. Primarily, tumours of an accessible size and whose upper limit does not pass the third sacral vertebra may be undertaken by the transperineal or parasacral route, as done in our study. Larger or very high (above S3) tumours may require abdominal or combined access.[1,3,7] On occasions it is even necessary to partially resect the sacrum to allow for suitable access to the surgical area or to carry out a block resection if it is infiltrated, which implies possible negative consequences such as incontinence and/or sexual dysfunction.[1,3,4,7,16,17] Consequently, we operated on 9 tumours by perineal route, 8 by abdominal, and 3 by combined route; resection with expected recovery was achieved in 85%. It is important to emphasize the need for optimal management of lesions which, ideally, in the case of malignancy, should be done by a multidisciplinary team with experience, comprised of colorectal surgeons, neurosurgeons,

vascular surgeons, traumatologists and plastic surgeons, along with medical oncologists and radiotherapists.[3,18]

Patients subject to complete tumour resections have presented varying survival rates; logically, prognosis of benign lesions was much better than malignant ones.[19] Recurrence of benign tumours ranges between 0% and 15% in the most extensive studies.[1,4,10] In our case, only 1 (7%) patient has presented with recurrence to date, after excision of a schwannoma. In the Mayo Clinic, the average survival rate at 5 years was 75%.[1] In our study, survival is 90% after an average follow-up of 3.5 years, and only 1 patient with mucinous cystadenocarcinoma presented with an inoperable recurrence due to infiltration in multiple pelvic structures.

Patients who cannot receive complete tumour resection may occasionally benefit from adjuvant treatment with high doses of radiotherapy,[3] and in chordoma treatment, a free interval without local progression at 32 months was recorded, with significant relief of symptoms for those who received this treatment.[20] In addition, thanks to the availability of tumour irradiation and neoadjuvant chemotherapy, the care for some patients has improved notably.

Finally, functional results from our study have been good regarding benign tumours, as previous evacuation problems improved where they were present, and no postoperative incontinence, given that even the parasacral procedure never included the sphincter muscle section, the pelvic floor or pudendal innervation, which in one case had to be identified and preserved due to the neighbouring lesion.

To conclude, presacral and retrorectal tumours continue to be a little known condition, with a rareness which makes diagnosis late and difficult, which may be made apparent after clinical suspicion with new imaging techniques. Its surgical treatment by surgeons experienced in pelvic surgery allows for good results and few complications to be achieved, except in the case of malignant tumours, for which histological type and infiltration of neighbouring structures depends.

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