Original Article

Outcomes of Surgical Intervention for Retrorectal Tumor: Experience of Sixteen Patients in Chang Gung Memorial Hospital

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Key Words
Retrorectal;
Presacral;
Tumor;
Mass

Background. Tumors originated from the retrorectal space are a rare group of diseases with diverse entities. The clinical features and surgical management of retrorectal tumors are seldom documented.
Materials and Methods. Medical records of adult patients receiving resection for retrorectal tumors during 1995 and 2008 were collected. Those patients with locally advanced colorectal or gynecologic adenocarcinoma were excluded. The clinical course, pathologic findings, associated treatment, and outcomes were assessed.
Results. A total of sixteen patients were included in this study, and five of them were diagnosed with malignant tumors. Benign tumors induced subtle symptoms and signs in 6 of 11 patients, while pain and other obstructive symptom were significant in malignant ones. Pelvic magnetic resonance image (MRI) and computerized tomography (CT) ascertained the diagnoses in all patients (14/14) accepting the examinations. Resections through abdominal, perineal, and combined approaches were introduced in 13, 1, and 2 patients respectively. Complete resection was achieved in 10 of 11 benign tumors but in none of malignant tumors. Teratoma, developmental cyst, and schwannoma were most common retrorectal benign tumors; on the contrary, pathologic result of retrorectal malignancies were diverse. All patients with benign tumors were disease free, except one experienced recurrence. However, all cases with malignant tumors were suffered from recurred or residual disease; three of them died within one year, whereas only one lived with disease.
Conclusion. Retrorectal benign tumors are frequently asymptomatic, while retrorectal malignancies more commonly manifest pain and compression symptoms. CT and MR scans provide sufficient information for diagnosis and preoperative therapeutic plans. Benign retrorectal tumors can be cured through complete resection; however, outcomes of malignant tumors are generally unsatisfied because of scarcely complete resection.
[*J Soc Colon Rectal Surgeon* (*Taiwan*) *2010;21:87-94*]

Retrorectal tumors are rare disease with heterogeneous entities occurring in a potential space between the rectum and sacrum, which is defined by fascia propia of the rectum anteriorly, by presacral fascia overlying the sacrum posteriorly, and by the ureters and iliac vessels laterally. The dome of the space is the peritoneal reflection, and the floor is the pelvic floor muscles. Complex vessels, lymphatics, and nerves re-

Received: December 15, 2009. Accepted: April 22, 2010.
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side in loose connective tissues. It is an area with multiple embryologic structures, so it has the potential to develop heterogeneous groups of benign and malignant tumors from the embryologic remnants. The incidence of the tumors is estimated about 1 of 40,000 to 63,000 admissions in the tertiary referral medical institutes.[1,2] The obscure anatomic location with difficult approaches and with diseases of multiple possible etiologies challenges clinical physicians to make diagnoses and therapeutic plans. The initial symptoms and signs in most patients are non-specific or insignificant, while the digital examination provides clues of clinical diagnoses. Magnetic resonance image (MRI), and computerized tomography (CT) were proved of improving accuracy in diagnostic and preoperative evaluation, but their efficacy of promoting the disease outcomes remains equivocal.[3-8] Previous reports recommended the curative resection as primary treatment, although it was difficult to be accomplished in the cases with malignant tumors.[2,4,5,8-10] In this study, we retrospectively reviewed our cases accepting surgical management of retrorectal tumors during a 13-year period in our institute.

Patients and Methods

Medical charts of adult patients, receiving surgical resection for retrorectal tumors from December 1995 to January 2008, in the department of Colorectal Surgery were collected. The data was obtained from the computer and paper medical chart system in our Linkou institute. We excluded those patients with tumors not arising from this potential space, including advanced colorectal and gynecologic cancers. A total of 22 patients were identified, and six of them were excluded due to incomplete data. The clinical presentations, diagnoses, surgical intervention and outcomes were reviewed and compared between benign and malignant tumors. The diagnostic methods and operative approaches were dictated by the clinicians. All the patients were primarily treated with curative resection, and neoadjuvant therapy was not introduced in this series. The final diagnoses were histopathologically confirmed. Major complications encountered included irreversible nerve injury, infection deep to the fascia layer, requirement of intensive care, and hospital mortality. Follow-up was performed by outpatient visits, and the date of contact was considered to be the final date of information. Survivals were calculated using the Kaplan-Meier method. The chi-square and two-sample t test were respectively used for discrete and continuous variables to assess statistical significance between groups. Value of $p < 0.05$ was considered statistically significant.

Results

In this study, five males and eleven females, with the mean age of 45.9 ± 16.2 years old, were included. There were 5 malignant tumors and 11 benign tumors. The age between patients with benign and malignant lesions was not significantly different, with the mean of 42.8 ± 16.8 and 52.8 ± 14.0 years, respectively. The mean follow-up peroid in these patients was 51.2 ± 49.4 months.

Symptoms and signs were summarized in Table 1.

Table 1. Comparison of demographics, presentations, and diagnosis between benign and malignant tumors

	Benign (n = 11)	Malignant (n = 5)	*p*
Demographics			
Mean Age (Year)	42.8	52.8	0.486
Sex (Male:female)	2:9	3:2	0.094
Clinical Presentation			
Symptoms			
Abdominal pain	1	4	0.005
Bowel habit change	3	2	0.611
Urinary symptom	2	1	0.931
Dysmenorhea	4	0	0.119
Palpable masses	4	1	0.513
Asymptomatic	6	0	0.037
Signs			
Digital examination	6	1	0.197
Lower extremities signs	1	2	0.142
Duration of Symptoms/ Signs (Month)	8.9	1.6	0.036
Diagnosis			
Diagnostic Tools			
Plain Film	0/2	0/2	
LGI series	1/3	1/2	
Colonoscopy	0/3	0/0	
CT	8/8	4/4	
MRI	4/4	2/2	
Diagnostic Tools Introduced	1.8	2.0	0.703

CT: computerized tomography, MRI: magnetic resonance image

Nearly half of patients with benign tumors (6/11) were incidentally discovered as an asymptomatic mass on digital or pelvic examinations, while all patients with malignant tumors came with distressing symptoms. Pain was the major initial presentation in our patients with malignant tumors, affecting 4 of 5 patients. The pain was located in the low abdomen in 4 cases, and involved in low back and lower limbs in 2 patients. Symptoms relevant to organ compression included urinary irritability, change of bowel habits, and sciatica and weakness of lower extremities, which were commonly associated with the malignant tumors than the benign tumors (1/5 versus 2/11; 2/5 versus 3/11; 2/5 versus 1/11); however, the difference was not statistically significant. Regarding physical examinations, benign tumors can be palpated in the fields of abdomen and perineum in 4 patients, while only one malignant tumor was able to be discovered through physical palpation over the abdominal area. In digital examination, benign and malignant tumors were palpated in 6 and 1 patients, respectively. The duration of symptoms from initial presentation to the diagnosis in benign tumors ranged between 0.5 and 24 months (average = 8.9 ± 7.2 months), whereas that in malignant tumors was shorter, 1.6 ± 1.1 months in average (range: 3 days – 3 months).

Results of image studies in our series were shown in Table 1. Low gastrointestinal series (LGI) illustrated the compressed luminal contour of the distal colonic segment, suggesting external structure, in 2 of 5 patients. Plain abdominal film failed to demonstrate the internal calcification as well as the external bony destruction in 4 patients accepting the assessment. Colonoscopy was unable to detect the intraluminal disease in 3 patients undergoing the examination. Nearly all the tumors in our series were identified by preoperative CT or MR scans, and the image reports were compatible with the surgical findings thoroughly. For example, in case No. 13 (Fig. 1), being diagnosed of chordoma, the infiltrative sacral plane discovered in MRI corresponded to the invaded periosteum disclosed in surgery. In case No. 11 (Fig. 2), being diagnosed of angiomyxoma, the caudal extension of the tumor mass to the lower pelvic region on images offered clues to determine the posterior surgical approach in advance. In case No. 7 (Fig. 3), being

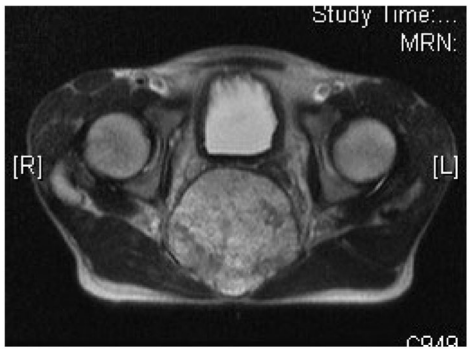

Fig. 1. Chordoma (patient no. 13). MRI demonstrated a voluminous, sizing 15*14 cm, mass located posteriorly to the rectum. An infiltrating border with the periosteum, along with a well-defined plane in its anterior and lateral sides, was observed.

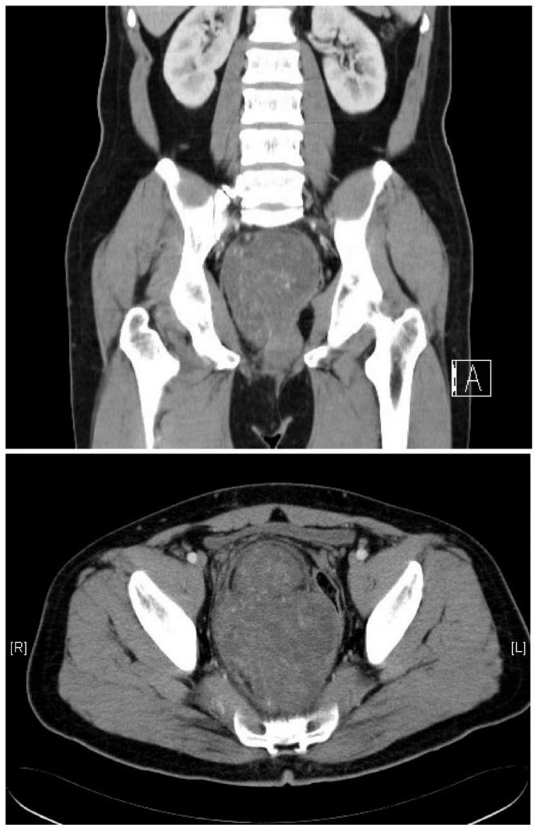

Fig. 2. Angiomyxoma (patient no. 11). Contrasted-enhanced axial and reconstructed CT showed a soft bulky, with multilobulated contouring, angiomyxoma located in the pelvis.

diagnosed of schwannoma, the origin of the tumor was clearly identified by the preoperative CT.

Abdominal, perineal, and combined approaches were performed in 9, 1, and 1 patients with benign tu-

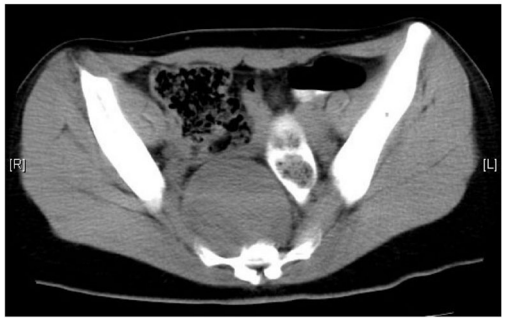

Fig. 3. Schwannoma (patient no. 7). Contrast-enhanced axial CT showed a solid and heterogenous mass that originated from the right sacral foramen and extended into the pelvic cavity.

mors, respectively, whereas abdominal and combined approaches in 4 and 1 patients with malignant tumors. The diameters of benign and malignant tumors were 7.1 ± 3.2 and 12.2 ± 2.5 cm in average, but the intergroup difference was insignificant. Curative attempts were undertaken in all our patients, except three patients with malignant tumors (1 chordoma, 1 GIST, and 1 malignant teratoma) in extensive local disease that restricted gross free resection margins. The durations of hospital stay for patients with benign and malignant tumors were between 4 and 26 days, and between 4 and 90 days, respectively, with 13.8 ± 5.8 and 32.0 ± 33.4 days in average.

The pathological findings of our patients were presented in Table 2. The most common etiology was related to teratoid differentiation, including one male patient with malignant disease and three female cases with benign entities (cystic components were discovered in two patients). Patients with benign teratomas were generally younger (age = 29, 47, 57 years old) comparing to whom with malignant teratoma (age = 77 years old). Developmental cyst and schwannoma were also common in benign groups, while pathology of malignant tumors presented diverse.

One intraoperative complication occurred in the benign group and three post-operative ones occurred in malignant group. The intraoperative complication was transaction injury of the sacral nerve as dissecting the severely matted schwannoma. The function of the nerve was reserved with mild sequel of sensory impairment after it being repaired by the plastic surgeon. Postoperative retroperitoneal and pelvic abscess developed in one patient after incomplete resection of epitheloid-sarcoma, he died of the uncontrolled local

Table 2. Demographic data of 16 patients

No.	Age	Sex	Pathology	Duration of Symptoms (m)	Diameter (cm)	Operative Approach	Hospital Stay (day)	Complication (s)	Follow-up (m)
Benign tumors									
1	27	F	Developmental Cyst	18	10	Anterior	12	Nil	88
2	68	F	Developmental Cyst	0.5	4.5	Anterior	19	Nil	171
3	41	F	Developmental Cyst	7.5	4.5	Anterior	26	Nil	128
4	57	F	Teratoma	24	8.7	Combined	12	Nil	60
5	29	F	Teratoma	2.5	12	Anterior	16	Nil	23
6	47	F	Teratoma	6	4.3	Anterior	9	Nil	22
7	24	F	Schwannoma	12	6.7	Anterior	15	Nil	83
8	21	F	Schwannoma	9	5.9	Anterior	4	Nil	82
9	69	M	Schwannoma	0.5	5.9	Anterior	12	Nerve Injury	25
10	46	F	Lipoma	6	5.4	Anterior	10	Nil	22
11	42	M	Angiomyxoma	12	12.7	Posterior	17	Deep Wound Infection	21
Malignant tumors									
12	48	F	Plasma Myeloma	0.1	13.3	Combined	90	Paraplegia, CNS. Infection	3*
13	48	M	Chordoma	1	14.4	Anterior	4	Nil	72
14	50	F	GIST	2	12.8	Anterior	19	Nil	15*
15	77	M	Teratocarcinoma	3	12.6	Anterior	25	Lung	1*
16	41	M	Epitheloid Sarcoma	2	7.9	Anterior	22	Lung, OP field Infection	3*

GIST: gastrointestinal stromal tumor, CNS: central nervous system, OP: operative, *: death in follow-up

infection as well as severe pulmonary infection 3 month following resection. Paraplegia and meningoencephalitis occurred in one case with plasma cell myeloma, who died of uncontrolled central nervous system (CNS) infection 3 months after operation. Severe lung complications occurred in the patients with epitheloid sarcoma and teratocarcinoma, and they died one and three months after surgeries.

All patients with incomplete resection had recurrent or residual diseases, including one of benign tumor and all of malignant tumors. Three patients with malignant tumors were followed by salvage therapy. Of them, the one with chordoma was alive while another with GIST accepting Gleevec treatment died of progression disease and the relevant medical illness 15 months after operation. The other one with incompletely resected epitheloid sarcoma received salvage radiation in total 4400 cGy/22 fx, but died 3 months later. The rest of two patients without adjuvant treatment died with residual disease in one and three months after operation. Patients with benign tumors were alive in regular follow-up. The only recurrence tumors was one case with schwannoma, which reappeared 4 years after the initial excision, and repeated surgical resection by the neuro-surgeon was performed in disease-free condition.

Discussion

This retrospective study reviewed outcomes of surgical intervention for retrorectal tumors during 13 years in our institute. These tumors are extremely rare, and the true incidence is difficult to be estimated. According to the prior literature, the majority of them were sporadically presented in the form of case reports. Several studies collecting more cases may provide information of the true incidence. Approximately 1.4 to 6.3 patients would be diagnosed of this disease on a yearly base in these major referral centers.[2,9-12] Though retrorectal tumors are scarce, they should be taken into consideration for those who present a posterior mass on rectal examination. Adult retrorectal tumors mainly occurred in female, aged 40 to 60 years, while malignant ones developed frequently in males.[2,4,5,8-10] Our data and prior reports were compatible. Though, the small number of patients may compromise the power of information; nonetheless, the diverse age distribution of cases regardless of benign or malignant tumors may imply the multi-etiologies as well as relative rarity of the disease.

Retrorectal tumors can be completely asymptomatic throughout their courses. The presented symptoms are usually related to compression of or invasion to the surrounding pelvis viscera and nerve. Urologic symptoms, change of bowel habits, and nerve root signs are common. Sean C. et al. compared clinical and pathological results of large series ranging from 1975 and 2004, with an overall of 298 patients.[5] They concluded that up to half of benign tumors were asymptomatic on diagnoses, while merely 4-14% of malignant tumors were clinically silent. Pain, bowel habit changes, and lower extremity signs affected 77, 51, and 25 percent of patients with malignant tumors. The results in our series were similar. More than half of patients with benign tumors had indolent clinical courses (6/11), while those who had malignant tumors presented pain, rectal compression and neurologic symptoms. The symptoms in our malignant tumors were positively related to the tumor size as well as local invasion. For example, intractable low back pain and lower extremity nerve root signs, being related to nerve invasion confirmed by surgeries, occurred in two patients with malignant tumors.

The awareness of the disease is critical for diagnosis; meanwhile, a careful rectal examination is the key, which provided the diagnostic rate of at least 39% of cases in previous reports.[4,8] In our study, seven patients (7/11) had positive findings in the rectal digital examination. An average of 1.9 diagnostic tools was introduced to each patient. Colonoscopy can examine the rectal lining and delineate the proximal extent of the tumors. It did not demonstrate intraluminal abnormality within three patients receiving the study, but it excluded the most common primary colon cancers. Lower gastrointestinal series can illustrate external compression or direct invasion upon the colon, which demonstrated rectal luminal filling defects in two of five patients undergoing this examination. However, the methods discussed above only provide indirect evidence of the disease. CT and MRI provide excellent direct anatomical description regarding the

lesions.[3-7] The images of CT with the conventional apparatus or the spiral single stratum apparatus are inferior to MRI because MRI provides the sequences on the sagittal and coronal planes, which reveal the origin of the lesions as well as their relationship with the surrounding structures more accurately than the images of the spiral CT does. However, CT with the multislice apparatus (16 strata) can yield results comparable to MRI with 1.5 T – 32 m T apparatus.[3] Thus, both CT and MRI techniques are optimal to determine the nature of the lesion and to plan the most feasible surgical approaches in advance. Our results were similar. Both CT and MRI represented the sensitivity of 100%, and the derived anatomical information, such as the infiltrating extension to surrounding tissues and the tumor origin, were compatible to all of our surgical findings. PET/CT (positron emission tomography/computed tomography) is a novel imaging device, which provides both functional as well as anatomical information of the target lesions. PET/CT detecting local recurrence of colorectal and gynecologic malignancy in retrorectal space started from 1990's.[13-15] However, its efficacy in diagnosing primary retrorectal tumors is still unclear.

All the tumors in this space should be excised completely.[2-5,7,8] Anterior, posterior, and combined approaches were alternative techniques depending on the size, location, and spatial relationship of the tumors.[7] For small (< 1 cm) and low-lying tumors, within the level of sacral promontory, posterior approach is favored. Anterior approach is preferred for larger, high-lying, or high-grade malignant tumors because it provides better visualization of pelvic structures, bleeding control, and easier mobilization of rectum. The average sizes of the benign and malignant tumors were 7.1 and 12.2 cm. In addition, three of five patients with malignant tumors were identified of perifocal invasion preoperatively. Hence, the anterior approaches were selected primarily in almost all our patients due to their greater size. The posterior approach was applied in one patient with angiomyxoma because of its posterior-inferior extension and low-lying position, and the combined approach was carried out in two tumors respectively due to the caudal extension, and extensively sacro-coccygeal involvement. There was no intra-operative shift of surgical approaches in our patients.

Iatrogenic surgical damage to surrounding structures includes urine/fecal incontinence, nerve root trauma, structural defect, and abscess/fistula formation.[2,4,5,7,8] The high complication rate was noted in patients undergoing sacrectomy. Neurogenic bladder and fecal incontinence occurred in 8-15 and 7 percents of patients undergoing such extensive procedures, and sometimes myocutaneous flap was indicated to the subsequent wound complications.[2,9] In our patients, sacrectomy was not performed, but nerve injury happened in two patients during dissecting their sacral planes. One received immediate nerve repair by plastic surgeon without severe neurologic sequelae, while the other patient had life-threatening CNS (central nervous system) infection. The incidence of abscesses and fistulae formation was reported high in patients with infected tumor contents and iatrogenic rectal injures.[2,16,17] In our study, one patient complicated operation field infection (retroperitoneal and pelvis abscess, in one with sarcoma), but intra-operative bowel injury or infective contents was not encountered in this case. The infection source of this patient was not determined.

The prognosis of patient with benign tumors receiving complete resection is excellent; however, which of malignant tumors is poor due to difficulty in performing safe resection margins.[2,4,5,8] In general, the 5-year and 10-year survival rates of completely excised malignant tumors were merely 40.7-75%, and 40-50% respectively.[5] The mean follow-up period in our series was 51.2 ± 49.4 months. Patients with benign tumors following complete resection were all in disease free condition, yet only one patient with malignant disease survived with disease (chordoma), who received radiation therapy for local recurrence and was alive with disease six years after operation. The rest of four patients with residual malignant diseases died in the 1st, 3rd, 3rd, and 15th month after operation. Our result supported the concept that disease free survival can be expected in benign tumors following curative resection, and generally poor outcomes were observed in malignant tumors.

To date, the role of radiation on managing the tumors was still under debate. Literature of preoperative/neoadjuvant radiation for retrorectal tumors was

not available. Adjuvant radiation for local residual and recurrent disease control remained controversial. Taking the most common retrorectal malignancy, chordoma, for example, Hobson et al. concluded that it was "notoriously resistant to radiotherapy" in a review article.[4] In Dis Colon Rectum, published in the same year, Glasgow et al. reported in another review article,[5] "aggressive postoperative radiation therapy has been reported for sacrococcygeal chordomas with favorable results." In our experience, the only survived patient with malignancy was diagnosed of chordoma which recurrence was effectively controlled by aggressive radiation therapy. The discrepancies in differed studies suggested further studies of cases as well as subsequent data. Conclusively, lacking sufficient long-term follow-up data, surgical resection remains the mainstream of treatment, and radiation and chemotherapy play an equivocal role in such tumors.

This is the second largest case series of adult retrorectal tumors in Taiwan. We presented sixteen patients with retrorectal tumors undergoing surgical treatment in our institute during 13 years. The benign tumors in our series mostly were discovered as an asymptomatic mass in routine digital and pelvic examination, while malignant tumors more commonly manifested pain, compression or neurologic signs. Male and elder patients were related to malignant retrorectal tumors in our series, although these two factors were not statistically significant. CT and MRI provided reliable diagnostic information. Prognosis following complete resection was promising, but it was difficult to be accomplished in malignant tumors. None of the malignant tumors in our series were completely resected, which indicated devastating outcomes in these patients with residual/recurrent malignant diseases.

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原　著

直腸後腫瘤外科治療成果回顧：林口長庚 16 例的經驗回顧

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目的　發生於直腸後的腫瘤是一群組織病理上差異性極大的罕見疾病，其在臨床上的病徵與外科治療的角色甚少被討論與確定。

方法　本研究蒐集自 1995 至 2008 年間在本院接受外科治療的直腸後腫瘤個案，大腸直腸腫瘤與婦產科腫瘤並不被列入討論，吾人等分析其臨床上的病史、病理診斷、外科治療、以及外科治療的成果。

結果　共有 16 個案例被納入本研究，其中有 5 例是惡性腫瘤。良性腫瘤 11 例中有 6 例其症狀是極其不顯著，然而疼痛與壓迫症狀在惡性腫瘤的案例是顯著的。在 14 例接受電腦斷層與核磁共振造影檢查的結果顯示 100% 的診斷率。13、1、2 個病患分別採取腹部、會陰、腹部合併會陰切除治療。畸胎瘤、發生囊腫、與許旺氏瘤是爲數最多的良性腫瘤，惡性腫瘤在本系列的病理診斷則差異極大。除了一個發生局部復發，所有的良性腫瘤都達到根除性外科治療的目的；相對的，所有的惡性腫瘤的個案都遭遇殘存或復發性疾病的問題，5 個中有 3 個於 1 年內死亡，僅有 1 例存活至今。

結論　直腸後良性腫瘤臨床多以無症狀表現，相對的惡性腫瘤較常以疼痛或壓迫性徵候來表現。電腦斷層與核磁共振造影對診斷與術前計畫可提供充足有效的臨床資訊。良性腫瘤一般可藉根除性切除達到治癒的目的，惡性腫瘤的預後普遍不佳，因爲根除性切除在這些腫瘤是很不容易的。

關鍵詞　直腸後、薦骨前、腫瘤、腫塊。