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Surgical management of retro-rectal tumors in adults

Abdellah Moufid (1,2), Mssrouri Rahal(1,2)Mdaghri Jalil (1,2)
khalidlahlou(1,2)Benamar Said (1,2), SettafAbdellatif(1,2),
1: Digestive Surgical Department B, Ibn Sina University Hospital. Rabat, Morocco
2: Faculty of Medicine and Pharmacy. Mohammed V University in Rabat. Morocco.
Corresponding author
Doctor AbdellahMoufid
Digestive Surgical Department C, Ibn Sina University Hospital. Rabat, Morocco

Abstract:
Introduction: *Retro-rectal tumours are a heterogeneous and diverse group of lesions that originate in the pre-sacral space. Their complex anatomy and difficult surgery make their management a real challenge.*
Materials and methods: *Our work is a retrospective study, spread over a period of 20 years, of a series of eight cases referred for retro-rectal tumors to the visceral surgery department B at the Ibn Sina hospital in Rabat, . Our series consists of two epidermoid cysts, a leiomyosarcoma, a fibromatosis, a hemangiopericytoma, a schwannoma, a ganglioneuroma, and a cystic rectal duplication. The aims of our surgical strategy are to study the different diagnostic means, therapeutic attitudes and their outcomes.*
Results: *There is a slight female predominance (62.5%), the median age is 43.5 years, 87.5 % of our patients were symptomatic. All masses were accessible to digital rectal examination. MRI and CT scan allowed identification of the masses and guided therapeutic management. The approach is discussed according to the size, nature and location of the mass in relation to the middle of the third sacral vertebrae. Four patients were approached via the perineal route, the abdominal route was performed for the others. The post-operative complications of this surgery seems to affect the quality of life of some patients. Complete resection limits recurrence and improves the prognosis of these lesions.*
Conclusion: *Retro-rectal surgery is both complicated and propitious to huge per and post-operative complications, so further training and surgical expertise are required for a careful approach, therapeutic success, and optimized outcomes*

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I. Introduction:

The tumoral processes occurring in the retro-rectal space constitute an extremely rare and heterogeneous entity, they bring real diagnostic and therapeutic challenges to the practicing surgeon, by their complex anatomical situation, as well as their difficulty of management. These lesions are a clinical rarity, with an estimated incidence of 1 in 40,000 up to 63,000 admissions to major referral centers [1, 2]. Many tumors remain asymptomatic for a long time, making their exact prevalence unknown. Retro-rectal tumors are congenital in two-thirds of cases, often asymptomatic, and generally discovered incidentally. Women seem to be more affected than men, however, malignancy is often found in males. The histological nature of the tumor, observed a priori at the time of surgery, can be identified preoperatively by biopsy according to radiological and clinical observations.

II. Methods:

Our retrospective study spans a period of 20 years, from January 2002 to December 2022, covers eight patients collected for retro-rectal tumors

The support of our study is made of observation which each includes: A clinical examination, possibly neurological and proctological, radiological assessments with in particular a CT and MRI examination, an anatomo-pathological analysis, and a post-therapeutic follow-up.

Its objective is:

o Study the main diagnostic and paraclinical parameters
o Study the different surgical approaches in terms of efficiency;
o Distinguish the evolutionary and prognostic profile of the different

III. Results:

The median age of our series of eight cases was 43.8 years with extremes ranging from 25 to 69 years.

Five cases were female; i.e. 62.5% of our study.

Medical and surgical history:
-Only one patient was a smoker.
-Only one patient had a gynecological history.
- All patients received their initial care. None were recurrent

The histological nature was distributed as follows:
-Three of our patients had congenital tumours: two epidermoid cysts and a cystic rectal duplication; all three masses are benign in nature. Congenital lesions represent more than 1/3 of our series compared to the study by Jao et al. where they represented two-thirds [1].
-Two patients had benign neurogenic tumours: Schwannoma and Ganglioneuroma.
-Three masses of various histological nature: Fibromatosis, Hemangiopericytoma and Leiomyosarcoma.
Our series did not report any bone lesions.
The malignant nature was found in two patients, i.e. 1⁄4, with a sex ratio of 1, and an average age of 43.5 years (42-45 years).

Symptoms: Pain is the main circumstance of discovery. All our patients were symptomatic, except for a single patient in whom the disease was discovered incidentally during a CT scan.
Most patients reported: chronic pelvic pain, urogenital disorders (pollakiuria, dysuria, dyspareunia) and intestinal disorders (constipation, tenesmus).

Digital rectal examination made palpation of each mass accessible, at least by its lower pole. It has a sensitivity of 100%.
All tumors were found by routine digital examination.
Palpation is the most effective, simple and economical method to identify these tumours. Proctoscopy is useful for large lesions, but usually leads to negative results for small tumors [1].

CT and MRI made it possible to give the different parameters of the masses by determining their size, location and their extension towards the adjacent structures. They thus predicted the suspected etiology and guided the operative strategy.

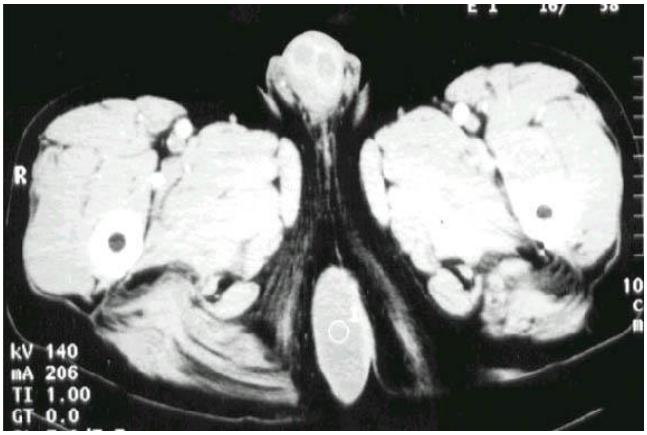

Fig 1:CT cross-section highlighting the right para-anal tumour.

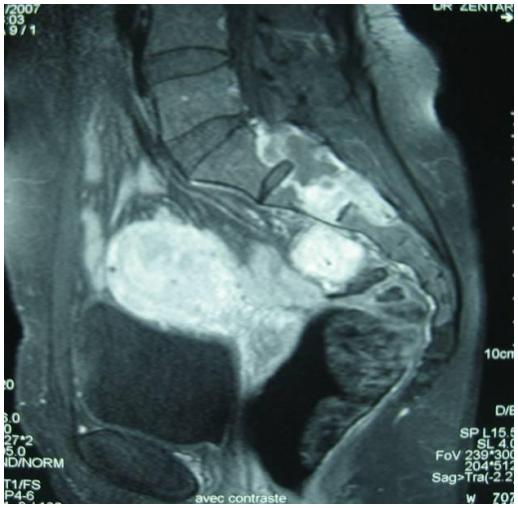

Fig 2 :**Coupe sagittale d'une IRM mettant l'envahissement du sacrum et du fascia pré-sacré par la tumeur**

The non-resectable nature of the mass in one patient raised the indication for a (scanno-guided) biopsy, which did not conclude with an exact histological profile.

A complete resection was required in all our patients, half of the cases were approached by the perineal route while the other by the isolated abdominal route.

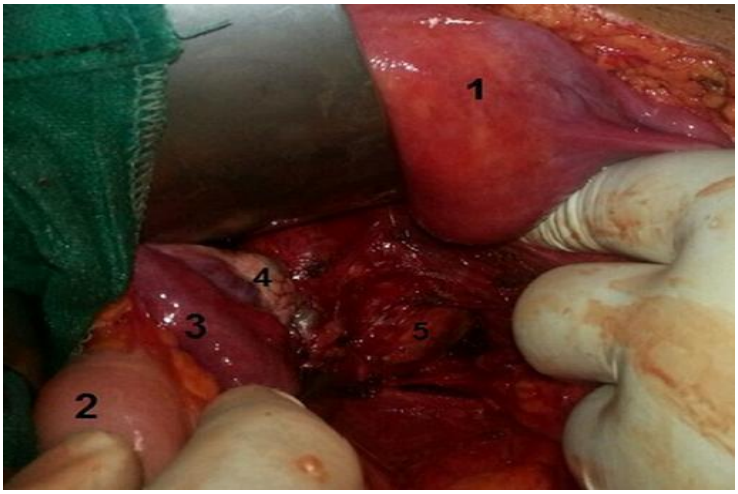

Fig 2 :Operative view highlighting the relationship of the mass with the adjacent organs; 1: Rectum, 2: Colon; 3: Uterus; 4: Ovary; 5: Retro-rectal mass.

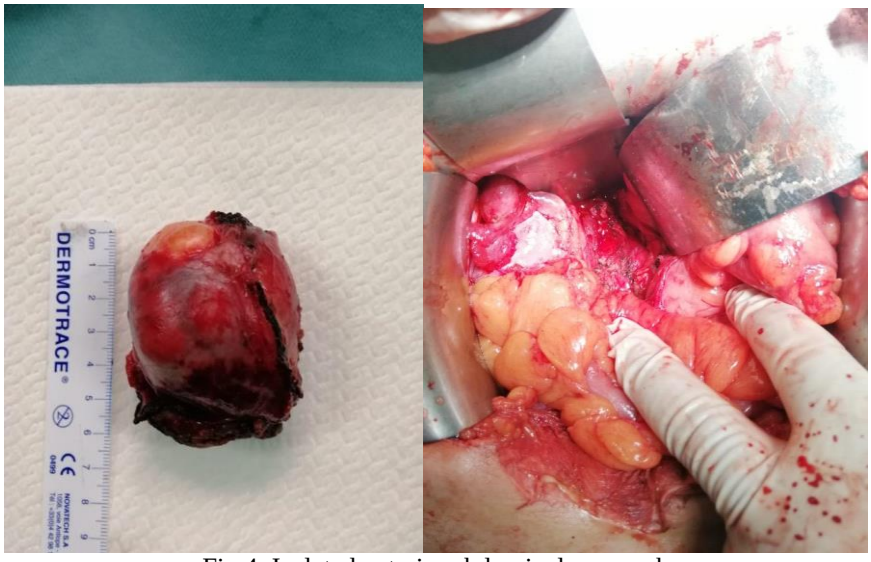

Fig 4: Isolated anterior abdominal approach

IV. Discussion:

The retro-rectal space is a site where several complex fetal changes occur. About 1/40,000 admissions turn out to be a retro-rectal tumor, according to 'Ewing': 1 in 35,000 newborns, and according to statistics from 'The Mayo Clinic', in the period between 1922 and 1936, 1 in 40,000 admissions presented with a retro-rectal tumor. Indeed these tumors are quite rare, 26 to 50% of these tumors are asymptomatic, thus making their real incidence difficult to interpret [3,4].
The incidence in our department is estimated at 0.0037%, or 1 case per 27,000 admissions. It is thus higher than in the literature.

Dziki et al. in 2016 [3], reported in a study including 29 patients, 16 male and 13 female cases, with a mean age of 48 years (19-80).
Jao et al. in 1985 [1], carried out work on 120 cases, with 60% women and 40% men, and reported an average age of 43 years (0-81).
Lev-Chelouche et al. (2003) [5], with a series of 42 patients, noted a sex ratio of 2 women to 1 man, with an average age of 41 years (21-84).
Glasgow et al. in 2005 [6], following a series of 34 cases, noted 21 female and 13 male cases, with an average age of 48 years (21-81).
Our series reports 5 female and 3 male subjects, with an average age of 44 years (25-69)

Malignant tumors represent on average 30% of retro rectal tumors in the large series reported in the literature. Congenital and benign retro rectal tumors represent 55% to 81% of the invariable series reported. Vestigial cysts predominate in women, including dermoid, epidermoid, and mucus-secreting cysts. These represent the majority of benign tumors, ranging from 8 to 62% in various series, with dermoid and epidermoid cysts representing 3 to 22% of benign tumors [7].
Among malignant tumors, chordoma is the most common, with an average of 38%. Chordoma is a congenital, solid or cystic malignant tumor, which appears mainly in men between 50 and 70 years old. Neurogenic retro rectal tumors represent 15%, while those of bone origin represent 3%, and retrorectal tumors of inflammatory origin (granuloma) 2.5%. The other types represent the remaining 19%. About 15% of cystic lesions and 65% of solid retro rectal tumors are malignant.[7].
According to the literature, the most common pre-sacral tumor is congenital benign teratoma. However, our study did not confirm this fact. This discrepancy may result from the small size of our sample and the age range excluding patients under 25 years of age, in whom congenital lesions may have been diagnosed and radically cured at an earlier age[7].

Presacral tumors are usually diagnosed late due to the absence of specific clinical symptoms or their asymptomatic nature. About 26 to 50% of patients in the literature are asymptomatic, which was not found in our series. The pain is often dull and poorly localized. The presence of symptoms appears to be related to infiltration of adjacent nervous and vascular structures. Infiltration of the sacral plexus will thus cause radiating pain to the lower limb and gluteal region, but can also be caused by inflammation in the minor pelvis. Low back pain may be associated [8]

Patients often complain of sacral or perineal pain, sometimes incontinence, constipation, defecation disorders, dysuria, fever and/or rectal bleeding. In our series, chronic pelvic pain was found in 50% of our patients.
Dysuria, incontinence and defecation disorders may result from either infiltration or mechanical compression of the pelvic nerves

Digital rectal examination was performed in all our patients, with a sensitivity approaching 90%. Indeed, it makes it possible to locate the mass in relation to the anal margin, and to note the different characteristics of the tumor, namely: its consistency, its size, its limits and its edges. It also allows, on its own, to evoke the diagnosis [9, 6]

Vaginal examination: materializes the extension of a large tumor Proctoscopy: aims to exclude associated colorectal pathology and should be considered immediately after pelvic examinations. The anoscopy also allows to see an abscess or a diverticulum or even a fistula. In case of narrowing of the rectal lumen, the upper limit could be noted

A neurological examination: meticulous and detailed is to be recommended, in order to apprehend a possible infiltration of the lumbosacral sympathetic chain

Transrectal or endorectal ultrasound: Confirms the liquid, cystic or solid nature of the mass by specifying its location, volume, homogeneity or heterogeneity, as well as the various relationships with adjacent organs and tissues. Endorectal ultrasound also provides details on possible tumor invasion and the presence of locoregional lymphadenopathy [10].
Endorectal ultrasound coupled with rigid rectoscopy has high sensitivity in the diagnosis of retro rectal tumors [11].

CT is the first line diagnostic tool in the study of any abdominopelvic pain. It can detect an anomaly in the pre-sacred space, without always giving a distinction between the benign or malignant nature. Indeed, this distinction requires a combination with endoscopic ultrasound [12]. A homogeneous lesion is in favor of the benign nature of the retro rectal tumor. On the other hand, a non-encapsulated heterogeneous lesion that enhances after injection of a contrast product is highly suspicious of malignancy.
The scanner makes it possible to evaluate the relationship between the tumor and the surrounding organs: uterus, rectum, bladder, ureters, and plan any pre or postoperative radiotherapy.

MRI plays a predominant role in the diagnostic approach of retro rectal tumors. It provides indications on the histological type of the lesion and clarifies the choice of the surgical approach.
It discovers the structure and the solid or liquid composition of the mass, its location in relation to the middle of the S3 vertebra, and its anatomical relationship with neighboring structures [13]. It distinguishes between benign and malignant tumors, with a sensitivity of 81% and a specificity of 83% for the preoperative diagnosis of malignancy [14]. More effective than computed tomography for the characterization of retro rectal tumors, it provides excellent anatomical detail and good soft tissue contrast [15].

Preoperative biopsy: Preoperative needle biopsy is controversial, not being very contributory, it is associated with a misdiagnosis rate of up to 44%. However, some authors have recently suggested that it should be considered [16]. The argument being that some of the patients may present with Ewing's sarcoma, osteogenic sarcoma, neurofibrosarcoma or desmoid tumor and who may benefit from neoadjuvant treatment. Therefore, a preoperative biopsy can significantly influence the preoperative management strategy and may alter the extent and nature of surgical resection [17].

The diagnosis of malignancy can be made by tumor markers. Indeed, the levels of ACE (carcinoembryonic antigen) and alpha-feto-protein have a specificity vis-à-vis teratomas. Thus a rise in their values may correlate with the malignant nature of the RRT. An increase in ACE alone would witness an epidermoid cyst. They are often used in postoperative monitoring and help detect recurrences [17].

Surgery is the mainstay of the management of pre-sacral tumors because it makes it possible to establish a diagnosis, prevent malignant degeneration and avoid secondary bacterial superinfection.
Its objective is to remove the pre-sacral lesion in its entirety with a minimum of morbidity for the patient.
Incomplete removal of the lesion can lead to the reappearance of symptoms, thus increasing the threshold for local recurrence, and decreasing the survival rate from malignancy. [18]

The criteria for determining the most appropriate surgical approach are: the size and location of the RRT relative to the middle of the S3 vertebra [19] [16] [20], the characteristics of the tumor as assessed by the results of imaging and/or biopsy, and the possible extension to neighboring organs: the sacrum and the pelvic organs.
According to M. Barraqué et al. It would therefore be interesting to distinguish approaches into 4 types [18] [7]:
- The abdominal route: Tumors located above the middle of S3, benign or malignant, without signs of extension to neighboring organs.
- The posterior approach: chosen for tumors, benign or malignant, located below the middle of S3.
-The perineal route: for low-lying, benign and small tumours.
- The abdomino-sacral route (or combined route): is indicated for tumors located above the middle of S3, and/or when there is invasion of a pelvic organ or a sacral vertebra.

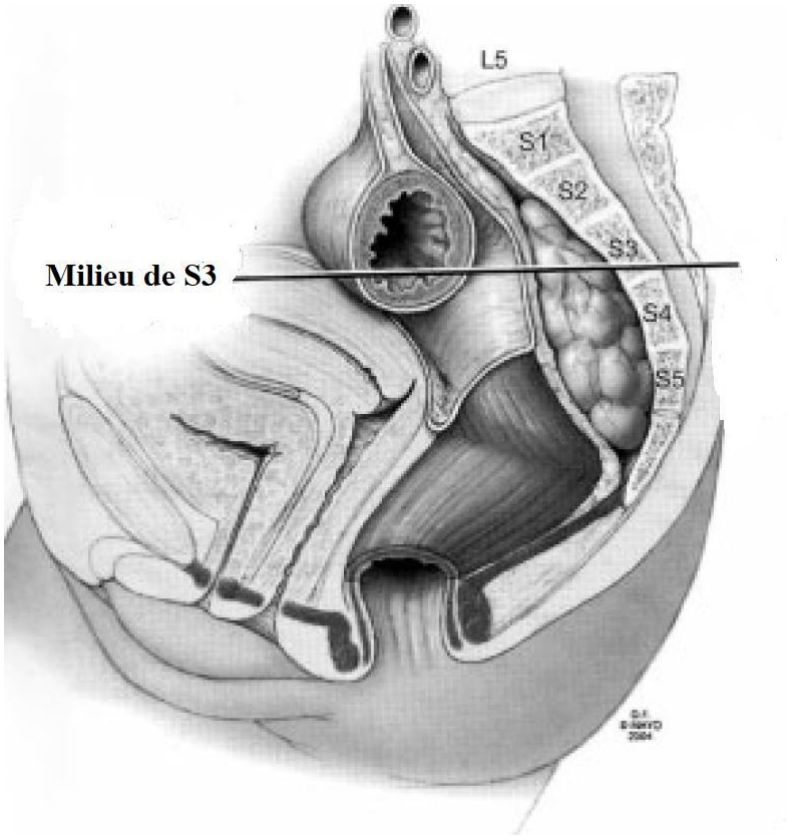

Fig 5 :Approaches according to tumor location [21]

The extent of the resection is determined by the nature of the pre-sacral lesion. Benign lesions can be removed by limited dissection, provided the lesion itself can be completely resected. Malignant lesions require radical resection, particularly if they adhere to adjacent structures, in which case en bloc resection of the lesion and the various elements involved (the pelvic vessels, sacral nerves, sacrum and rectum) becomes necessary [17].

Indeed, the resection of chordomas is more difficult than that of teratomas because of their propensity to invade adjacent structures. In case of large developmental cysts (> 4-5 cm), the abdomino-sacral (combined) route would be preferred because it can be difficult to judge the cephalic extent of a tumor lying in this tight space, by a posterior approach alone [16].

For tumors with proven sensitivity to radiotherapy and/or chemotherapy, a simple transabdominal excision (in the case of small neuroblastomas) or a generous biopsy for the study of tumor markers (in the case of lymphomas) can be adequate. Furthermore, the discovery of extensive chordoma in a patient with few symptoms, elderly or at low risk may encourage the surgeon not to perform excision at all or, in the event of an obstruction, to perform a palliative colostomy as the only alternative. [22].

The advantages and disadvantages of each of these approaches should be considered [23, 24]. MRI is the test of choice to select the appropriate surgical approach [25].

laparoscopy [26] can reduce the duration of the operation, the duration of hospitalization, the risk of recurrence, and offers better recovery with less postoperative pain [27]. It perfectly visualizes the pelvis and facilitates dissection compared to open laparotomy [28].

Robotic-assisted resection is a safe, effective and minimally invasive technique to approach benign pelvic tumors, with low morbidity and a 92% R0 complete resection rate. It can be proposed even for a tumor situation below S3, unlike abdominal laparotomy, and in front of a highly vascularized tumour, ligation of the median sacral artery makes it possible to avoid massive hemorrhage leading to cardiac arrest and dead.

Nevertheless, it is clearly preferable to ensure that the tumor is benign before recommending a laparoscopic approach [26]. Recently published studies suggest that adequate exposure and resection can be performed with laparoscopic access, particularly when malignancy has been ruled out [29].

Regardless of the choice of method, complete tumor resection with a large margin of healthy tissue is the primary goal of surgical management. The long-term prognosis depends on the histological nature and the quality of the surgical excision. It is an important criterion, which determines the overall prognosis, including survival, morbidity and prevention of recurrence or malignant degeneration of a residual fragment [7]. The absence of tumor recurrence highlights the effectiveness of a surgical procedure [3]

V. Conclusion :

Retro rectal tumors constitute a heterogeneous and diverse group. They include congenital or acquired, benign or malignant tumors.surgery with complete excision and respect for healthy margins allows therapeutic success to flourish. The surgical modalities are discussed according to the nature of the tumor, its location in relation to the middle of the 3rd sacral part, its size, its height and the involvement of adjacent elements. The main approaches are: Posterior, perineal, isolated abdominal and abdomino-posterior (mixed).

Adjuvant therapy is supported by histological type. The latter accompanied by the quality of surgical excision are the main indicators of long-term prognosis and survival rate.

Data Availability Statement

The data that support the findings of this article are available from the corresponding author upon reasonable request.

Competing interests:

The authors have no conflicts of interest and source of funding. The subject of the article had no commercial interest, no financial or material support.

Ethics statement

Drs AbdellahMoufid, , SettafAbdellatif ,Benamar Said , khalidlahlou ,Mdaghri Jalil , Mssrouri Rahal declare no conflict of interest.

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