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Transsphincteric tumor resection in case of a pararectal solitary fibrous tumor

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ABSTRACT

Transsphincteric resection of rectal tumors was first described about 120 years ago. Nowadays, this approach faded into obscurity due to standardized guidelines and practice in surgical oncology including lymphadenectomy, mesorectal excision and radical dissection of veins. However, transsphincteric resection seems reasonable in some cases, especially if an abdominal approach can be avoided.

In the following, we will present and describe the technique of the transsphincteric approach with its variations in rectal surgery in the case of a rare pararectal tumor.

1. Introduction

Access to the anal canal, the distal rectum and the mesorectum can be established by splitting the sphincter. Kocher and Verneuil described in 1875 the posterior exposure of the rectum through a coccyx resection [1]. One year later, Cripps described in a series of 36 patients access to the rectum through a transsphincteric approach. Despite the lack of sphincteric reconstruction, 2/3 of the patients did not develop anal incontinence. In 1885, Kraske modified the approach by gaining access through a pararectal incision with resection of the sacrum to avoid dissection of the sphincter [2]. In the following years, several publications confirmed the feasibility of this technique, however, the abdominal approach for the treatment of benign and malignant tumors in the pelvis became the 'gold standard' [3].

In 1974 York Mason modified the technique by reconstructing the sphincter after initial dissection with good results on postoperative anal continence [4].

Huber, von Hochstetter and Allgöwer reintroduced the anatomy and its clinical relevance of the pelvis and its relation to sphincteric reconstruction, which lead to the implementation of surgical technique, anatomical and physiological knowledge into today's surgery [5].

The dorsal transsphincteric approach is an applicable technique that allows the surgeon to operate on the distal rectum and mesorectum without opening the abdominal cavity.

2. Definition

The dorsal transsphincteric approach describes a surgical technique that allows access to the mesorectum, the distal part of the rectum and the anal canal by dissecting the levator ani and part of the sphincter through a parasacral incision.

3. Surgical technique

Prior to surgery the rectum should be mechanically prepared. The patient is then placed into the prone position. The incision should run parasacral, about 3 finger width above the coccyx, down to the anocutan line. The distance to the sacrum should be 1 finger width. Following the dissection through the subcutaneous tissue, the M. gluteus maximus is split at the cranial part of the incision displaying the sacrospinal ligament cranially and the external anal sphincter caudally. The musculature of the pelvic floor should be dissected medially sparing its innervation and perfusion. Depending on the location, size and type of the tumor, dissection of the sphincter musculature might be necessary. While dissecting the levator ani and the external and internal sphincter, it might be useful to ligate the musculature facilitating a later reconstruction.

Once the dissection is completed the Waldeyer's fascia can be identified and resected along with the tumor thus gaining access to the mesorectum. The rectum can then be mobilised along the rectoprostatic fascia. If the preparation needs to be extended cranially, dissection of the sacrospinal ligament is necessary.

Once resection is completed, the corresponding parts of the levator ani and sphincter can be sewn together. Gathering of the puborectal muscle can be discussed to prevent possible prolapse or incontinence.

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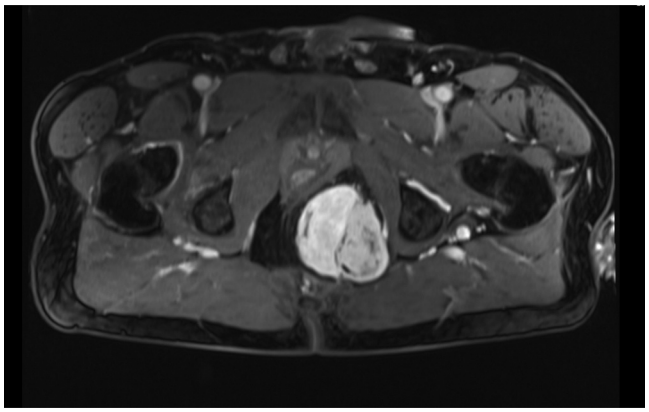

Fig. 1. T1-MRI-Scan: the tumour is hypervascularized in the Fossa ischioanalis.

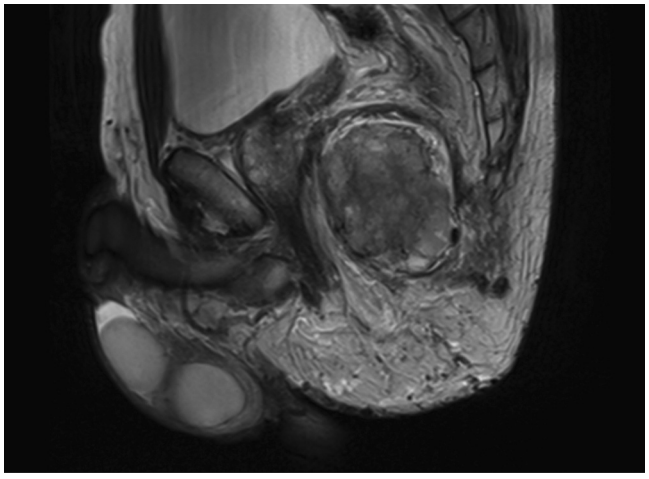

Fig. 2. T2-MRI-Scan: the tumour in sagittal projection in the mesorectal tissue.

4. Case report

We present the case of a 60-year-old male patient who received an abdominal computed tomography for unintentional weight loss of 3 kg. An upper endoscopy revealed a long-barret esophagus. A colposcopy was normal. Clinical examination revealed no pathological findings.

The CT of the abdomen and pelvis described a $9 \times 6 \times 6$ cm hypervascularized solitary tumor pararectal in the fossa ischioanalis. A MRT confirmed the presence of the tumor with tumor-supplying vessels from the A. iliaca interna. An infiltration of neighboring organs was excluded (Figs. 1 and 2).

We decided to embolize the tumor preoperatively followed by a transsphincteric tumor resection (Fig. 3). The surgical resection was performed as outline above. The tumor was extirpated followed by reconstruction of the pelvic floor. The rectum was not affected during the procedure.

The patient was discharged on the 7th postoperative day. During patient follow-up a superficial wound-healing deficit developed which was treated accordingly and healed secondary. The patient had no symptoms regarding anal incontinence.

5. Histology

The histopathological examination revealed a solitary and vascularized tumor with an immunohistochemical CD34-reactivity and a nuclear STAT6-expression. In addition 6% of the cell nuclei expressed the Ki-67-Antigen (Fig. 4). The tumor cells showed a positive reaction toward the Vimentin (Fig. 5) antibody with negative markers for the S-100 antibody. The results of the conventional histology showing a solitary vascularized tumor (Fig. 6), and the immunohistochemical analysis suggest the diagnosis of a solitary fibrous tumor.

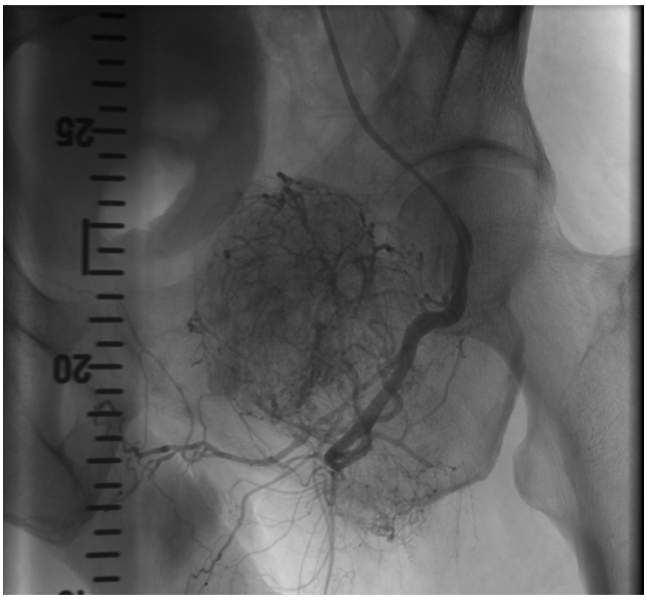

Fig. 3. Angiography: the tumour vessels origin at internal iliac artery angiography: the tumour vessels origin at interal iliac artery.

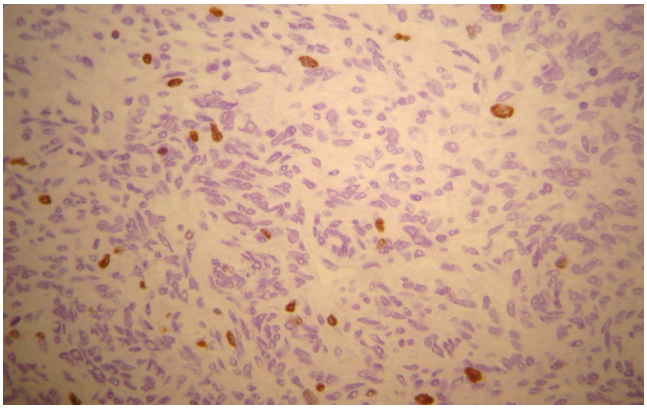

Fig. 4. Expression of Ki-67 antigen (6%).

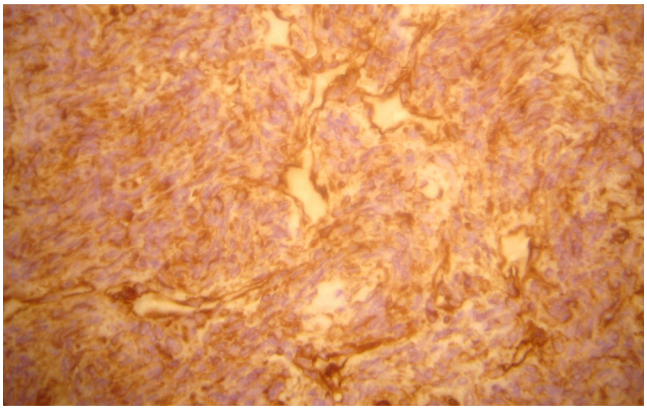

Fig. 5. Expression of vimentin.

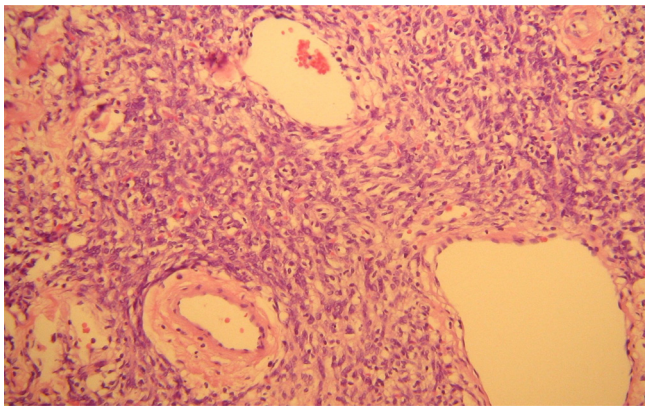

Fig. 6. HE-histology: well vascularized tumour.

6. Solitary fibrous tumor

The tumor entity was first described in 1931. Less than 2% of all soft-tissue-tumors belong to this group [6]. Most tumors of this type are benign, however, 10–15% are malignant and highly aggressive with the formation of distant metastasis. The micromophology of the tumor does not give relevant information about its growth characteristics [7].

For many years it was believed that solitary fibrous tumors are restricted to the pleura or the mesothelium. Recent studies regarding its histogenesis revealed a much more heterogenous picture, showing signs of fibroblastic and myofibroblastic origin [8].

For a long time hemangiopericytoma was used as a synonym, although the recent WHO classification defines hemangiopericytoma as a subgroup of SFTs [9,10].

The histopathological examination in combination with the immunohistochemistry is crucial for diagnosing SFT. The characteristic microscopic changes are described as “patternless pattern” with hypo- and hypercellular areas and so-called “staghorn” pattern with ectatic tumor vessels [11]. The additional information given by the immunohistochemistry allows further discrimination among the differential diagnosis such as sarcomas, metastases or GIST. Typical positive markers are CD34, Vimentin, bcl-2 and CD99. Contrary, markers such as S100, actin, SMA, CEA and keratin are typically not present [12,13]. Current analysis showed that a nuclear expression of STAT-6, as presented in our case, is a very specific marker for the presence of a SFT [14,15].

Malignant SFTs can present with specific features but without a related causality. Histopathological results do not suggest a specific biological growth pattern of SFTs. Generally, large tumors with atypical cell nuclei, more than 4 mitosis/10HPF, growth of necrotic areal and hemorrhages are criteria for a malignant potential. A retrospective analysis described a patient age >55 years, a tumor size >15 cm and a high mitotic index as risk factors for metastasis and a increased mortality [16]. Also benign SFTs have the potential to develop into malignancies. It was shown histochemically that the reaction to CD34 decreases while there is an overexpression of S100 and p53 [17]. It is also known that some parts of SFTs are well-differentiated while others are not. Molecular genetic analysis have shown that the mutation of p53 and NAB2-STAT6 influence the clinical growth pattern of SFTs [18,19].

SFTs of the small pelvis as described in this case are rare. Most SFTs present in the 5th decade and are usually asymptomatic. Once the tumor reached a certain size, causing displacement of adjacent organs, it becomes symptomatic. Symptoms range between diffuse abdominal pain, impaired bladder emptying and related neurological symptoms involving the sacral plexus. In addition, hypoglycaemia is possible due to tumors producing insulin-like growth factor [20].

With the knowledge of a potential malignant transformation, radical surgical tumor extirpation is the treatment of choice. The 5-year survival rate is close to 100% after a R0-resection [21]. Guidelines on safety margins do not exist. A follow-up is indicated, however, duration and frequency for follow-up visits are not defined.

There are some cases of recurrencies that presented many years after the primary resection of the tumor, indicating a long follow-up. In the case of recurrence or metastases, surgery remains the treatment of choice. The use of radiotherapy and chemotherapy (e.g., doxorubicin) has been described, however, no significant change in long-time survival was achieved [22,23].

In the case of non-resectable SFTs, Espat and others were able to show certain positive effects due to the implementation of antiangiogenetic therapeutics [21].

7. Discussion

This case demonstrates the possibility of a pararectal approach in extirpating a tumor of the small pelvis. Due to the tumor biology of a SFT, R0-resection is absolutely necessary to prevent later recurrence. Achieving a R0-resection determines the prognosis of the patient [24]. Extrapleural SFT can present across the body area, however, SFTs affecting the mesorectum are very rare. The histopathological and biological properties of pleural and extrapleural SFTS are similar [25]. Metastases can appear years later and should be surgically resectable, if possible.

A CT or MR should be performed to diagnose a SFT. A SFT cannot be safely differentiated from other tumors although some SFT-related characteristics such as hypervascularisation with hemorrhages, necrosis and cystic lesions within the tumor are described. A hypervascularisation was noticed in our case. Therefore, a preoperative embolization was performed. A MRT can identify a delineation or infiltration to adjacent structures and the sacral vertebrae.

A posterior approach for resection of the tumor is only possible if the tumor lies caudally to S3. Otherwhise an abdominal approach is indicated.

By means of this case we want to demonstrate the feasibility of a posterior pararectal approach in resecting a tumor of the small pelvis. This historic and neglected approach is indicated in some instances and prevents complications such as hernias, postoperative paralysis of the colon and injury to adjacent structures that might follow open abdominal surgery.

As we described above different types of posterior approaches were described in the past. The usual opinion of these approaches is that a transspincteric approach as described by Mason or modified by Allgöwer might result in an anal incontinence. This is not correct. In contrast to that it would by necessary to resect the coccys and sometimes part of the sacrum as described by Kraske. Therefore it may result a larger wound defect. Also some patients describe an unpleasant sensation after resection of the coccys.

That is the reason why we would prefer the transsphincteric resection and not the resection of coccys and/or sacrum.

R0-resection of SFTs is the treatment of choice. Preoperative embolization facilitates the constraint on tumor perfusion and related risk of intraoperative bleeding.

8. Summary

We present the case of a patient with a solitary fibrous tumor. A preoperative remobilisation of the tumor was performed followed by a radical R0-resection through a posterior approach thus sparing

an abdominal approach. The biological properties of the tumor cannot be predicted. An adjuvant therapy is not established, although late recurrencies are described in the available literature. Therefore, an intense patient follow-up is indicated.

Conflict of interest

None.

Funding

None.

Ethical approval

Because it is no experimental study and retrospectively analyzed there is no need for an ethical approval.

Author contribution

Achim Troja collected the data and wrote the manuscript. Nader El-Sourani translated the manuscript. Dalibor Antolovic participated in the surgery and gave ideas for publishing. Hans Rudolf Raab inspired us by his work and teach us in operative technique.

Guarantor

Achim Troja is the guarantor.

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