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Rare tumorous and pseudotumorous lesions of the Rectum and Perirectal Tissue

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Rare tumorous and pseudotumorous lesions of the Rectum and Perirectal Tissue

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Abstract

Aim:

Rare tumors of the rectum and perirectal tissue present diagnostic and therapeutic challenges due to their rarity, heterogeneity, and limited data in the literature. This study aims to share experiences from a large coloproctology center to improve the understanding of these tumors' therapy and outcomes.

Method:

In a retrospective case series, data from 15 patients with rare perirectal and rectal tumors who underwent rectal resection at the Department of Coloproctology at Prosper Hospital of Recklinghausen between 2010 and 2023 were analyzed. The study focused on the clinical and pathological features of these tumors and evaluated the impact of surgical resection on patient outcomes.

Results:

The study included 15 patients (10 women, 5 men) with a mean age of 58 years (range: 31 - 85 years). Most patients (n=12; 80%) underwent rectal resection with curative intent. Achieving an R0 resection often required multi-visceral resection (n=5; 43%). Procedures were mostly complication-free (n=9; 60%), resulting in recurrence-free survival for 9 patients (60%) over a mean period of 33 months (range: 5-60 months).

Conclusion:

Treating rare tumors of the rectum and perirectal tissue requires an interdisciplinary approach and individualized therapeutic regimens. Malignancies should be managed within the context of a tumor conference. In this study, surgical resection led to satisfactory outcomes for most patients.

Introduction

Adenocarcinoma, typically found in older patients, is the most common malignant rectal tumor, making up over 95% of cases [1]. When occurring in younger patients, it often has a genetic predisposition. Adenocarcinoma during pregnancy is rare (incidence of 1:13,000) and is one of the rarer malignant rectal neoplasms, with only a few hundred cases published [2]. Diagnosis is usually late due to non-specific symptoms like bleeding and changes in bowel movements being attributed to other proctological diseases [3-4]. Other malignant tumors in the rectum and perirectal tissue are uncommon and include lymphomas, sarcomas, neuroendocrine tumors, gastrointestinal stromal tumors (GIST), squamous cell carcinomas (SCC), metastases from other carcinomas, and masses with malignant ectopic tissue. These tumors differ significantly from rectal adenocarcinoma in terms of disease course, therapy, and prognosis. Due to their rarity, there are few publications discussing these tumors [5-16]. The hidden location in the lesser pelvis often leads to late-stage diagnosis if symptoms are non-specific. They are usually discovered accidentally during rectal or vaginal examinations in preventive measures and are increasingly noticed as incidental findings due to the growing use of sectional imaging in everyday medical practice.

As one of Germany's largest coloproctology clinics, Prosper Hospital of Recklinghausen presents its experience with rare tumors of the rectum and perirectal tissue in this study. We detail the clinical and pathological features of these tumors and assess the importance and extent of surgical resection, particularly regarding patient outcomes and prognosis.

Method

From January 2010 to March 2023, 60-70 rectal resections were performed each year in the Department of Coloproctology at Prosper Hospital of Recklinghausen. The patient population from this time period was assessed for the incidence of rare tumors of the rectum and pararectal tissue. Based on an evaluation using the ORBIS hospital information system, which records the clinical data of the treated patients, 15 patients were identified who underwent rectal resection for rare tumors of the rectum and pararectal tissue. There were 10 women and 5 men with a mean age of 58 years (range: 31 - 85 years).

During a retrospective workup, the clinical data from these patients was examined with regard to the following parameters: gender, age, symptoms, ASA classification, clinical course, results of preoperative diagnostics, preoperative suspected diagnosis, therapy, histological findings, postoperative complications, and postoperative outcome.

All patients underwent preoperative colonoscopy with rectoscopy and sampling, along with computer-assisted tomography (CT) of the abdomen and thorax. Further staging diagnostics were carried out depending on the respective tumor entity (Table 1). All biopsies were examined by a gastrointestinal pathologist (Pathological Institute of Recklinghausen and Institute of Pathology at Ruhr University of Bochum).

Wherever malignant tumors were involved, the therapeutic treatment plan was determined during the interdisciplinary tumor conference. Postoperative follow-up examinations were performed by the Oncology and Coloproctology department of Prosper Hospital of Recklinghausen.

A favorable opinion by the ethics committees of the Medical Association of Westphalia-Lippe and Westphalian Wilhelm University of Münster were obtained (file number: 2022-614-F-S from October 2022).

Results

History

In 12 (80%) of the 15 patients, the rectal tumor or the perirectal tumor was found during further diagnostics, which were initiated due to the existing symptoms. In one case, it was an incidental finding during a cesarean section. Two additional tumors were discovered during a screening colonoscopy and a follow-up tumor examination 6 years after gastric carcinoma. Apart from one patient (patient no. 13), all other patients suffered from non-specific symptoms: pain (n=13), hematochezia (n=6), stool irregularities (n=8), B symptoms with weight loss, fever, night sweats (n=5) (Table 1).

14 patients had a negative or unknown family history of colorectal carcinoma in first-degree relatives (family history negative (n=9), unknown (n=5)). One patient's first-degree relatives suffered from a malignant tumor (colon carcinoma).

With regard to secondary diseases, the following ASA levels were found for the affected patients: ASA 1: (n=3), ASA 2 (n=5), ASA 3 (n=7) (Table 1).

Diagnostics

Depending on the findings of the primary diagnostics (endoscopy with sampling and CT of the chest and abdomen), further examinations and/or interventions were initiated depending on the tumor characteristics (Table 1). There was tumor-related stenosis of the rectum in 7 patients (47%).

The diagnostics conducted led to the following suspected diagnoses: Metastasis (n=3), inflammatory tumor (n=1), SCC of the rectum (n=4), Douglas SCC (n=1), adenocarcinoma of the rectum (n=2), benign cystic perianal mass (n=1), giant presacral tumor of unclear malignancy (n=1), giant perirectal cyst with suspected dermoid cyst (n=1) (Table 1).

Based on the biopsies taken preoperatively, a clear diagnosis was made in 9 patients, which was also confirmed in the final histology of the rectal resection. In 2 patients, despite the absence of tumor cells in the biopsies removed preoperatively, there was clinical suspicion of malignancy, which led to local tumor resection and thus to the diagnosis. In 4 patients, the final diagnosis could not be made until postoperatively after rectal resection (Table 2).

Therapy

Surgery was indicated for all patients with the mentioned suspected diagnoses. Total mesorectal excision (TME) was performed in 10 patients (67%). In 4 cases (27%), discontinuity resection or rectal extirpation with a permanent stoma was done. One patient underwent transanal full-thickness resection.

One patient (no. 12) had intersphincteric rectal resection with a permanent stoma for palliative care due to perforated rectal carcinoma. TME was also done with palliative intent

in the patient with hepatic metastatic rectal carcinoma during pregnancy (no. 13) and in the patient with metastatic stenosing SCC (no. 8). All other surgeries were curative (Table 1).

In 5 patients (33%), an extension to multi-visceral resection was required: resection of the coccyx tip due to ectopic pancreatic carcinoma (patient 4), hysterectomy and peritonectomy in the lesser pelvis due to Douglas squamous cell carcinoma (patient 10), adnexectomy, hysterectomy, cystectomy, and nephrostomy for metastatic gastric carcinoma (patient 1), rectal extirpation with en-bloc cystectomy and ileal conduit due to poorly differentiated adenocarcinoma with rectal and bladder infiltration (patient 5), and hysterectomy due to rectal carcinoma during pregnancy (patient 13) (Table 2).

The preoperative suspected diagnosis was confirmed in only 9 of 15 patients (60%). In 6 patients (40%), tumor resection was needed to establish the diagnosis (Table 2).

Histological findings

Histological examination revealed malignancy in 14 patients (94%) (Table 2). Metastases were found in 3 cases: metachronous peritoneal metastasized gastric adenocarcinoma infiltrating the rectum 6 years after gastrectomy, prostate carcinoma metastasizing presacrally into the mesorectum 8 years after surgery and anti-androgen therapy (Fig. 1), and GIST recurrence in the retrovaginal space with liver metastases. The GIST was first diagnosed 35 years ago, with 6 local recurrences and multiple liver metastases.

Other malignancies included ectopic pancreatic carcinoma in the rectum without primary pancreatic involvement (Fig. 2), rectal adenocarcinoma with bladder infiltration, four instances of rectal SCC, one Douglas SCC with rectal infiltration (Fig. 3), perianal cystic mucinous adenocarcinoma (Fig. 4), and Fournier gangrene due to rectal cancer perforation during neoadjuvant radiochemotherapy (Fig. 5).

Two patients had rare rectal adenocarcinomas: one incidentally found during pregnancy with hepatic metastases discovered during a cesarean section, and another with a triple tumor in the rectum, sigmoid colon, and an 8 cm presacral lymph node metastasis (Fig. 6). One patient had a benign mass, an inflammatory tumor with rectal stenosis caused by graft-versus-host colitis 5 years after a bone marrow transplant (Table 2).

Postoperative Course / Follow-Up

The postoperative course was uneventful in 9 patients (60%). One 85-year-old (ASA 3) patient (no. 11) died 2 days after rectal extirpation due to pulmonary decompensation from perianal cystic mucinous adenocarcinoma. Local wound healing disorders occurred in 3 patients (25%). One patient developed an intra-abdominal abscess requiring surgical debridement and vacuum therapy (no. 1), while another had an abscess drained under CT guidance (no. 8) (Table 3).

After the interdisciplinary tumor conference, one patient was diagnosed with metastatic GIST and started on imatinib therapy. The patient with recurring prostate carcinoma received antihormonal therapy. Chemotherapy was initiated in 7 patients, and adjuvant

radiochemotherapy in one patient after transanal full-thickness resection for SCC, and another after TME for SCC (Table 3).

The mean postoperative follow-up was 33 months (range 5-60 months). Nine patients (60%) remained recurrence-free, while 3 patients (20%) had recurrences. Two patients (13%) died due to tumor progression (Table 3).

Discussion

Because of ongoing developments with diagnostic procedures, particularly imaging procedures, along with the increasing use of screening colonoscopies in addition to adenocarcinoma of the rectum, rare tumorous masses adjacent to the rectum or within the rectum are often detected as incidental findings. With the majority of these tumors, the data on diagnosis, therapy, and prognosis is very poor due to their rarity and heterogeneity. In this paper, we use a case collective to report on our experiences with 15 patients with rare tumors of the rectum and pararectal tissue who underwent rectal resection in the Department of Coloproctology at Prosper Hospital of Recklinghausen between 2010 and 2023.

Despite exhausting all means of modern diagnostics and sampling, preoperative confirmation of the diagnosis was only possible in 9 patients (60%) in our patient collective. All patients underwent resection of the tumor-afflicted or surrounding portion of the rectum. The operation was performed with curative intent in 12 patients (80%) and with palliative intent in 3 patients (20%).

Distant metastasis was the cause of 2 additional tumors with infiltration and walling of the rectum. A 58-year-old patient (patient no. 1) exhibited metachronous gastric carcinoma metastasis in the rectum. The initial diagnosis was made 6 years previously. At that time, the patient had been treated curatively with neoadjuvant chemotherapy and subsequent oncological gastrectomy. En-bloc rectosigmoid resection with hysterectomy and adnexectomy was then performed. Chemotherapy was administered postoperatively. Bladder and rectal metastases were discovered at the follow-up (6 months postoperatively). TME was then performed with supra-anal blind closure, mucous fistula of the neosigmoid colon, cystectomy, and nephrostomy. The patient lived recurrence-free until the end of the following follow-up period (48 months).

Metastatic spread to the rectum is a rare finding, and the management of rectal metastases is by no means standardized [17-18]. Surgical treatment appears to be a valuable treatment option for certain patients, while the overall prognosis depends mainly on the primary tumor [17]. In this context, Janjic et al. published a systemic review of the literature in 2022 based on 99 publications with a total of 162 patients with rectal metastases from 16 different primary tumors. The most common origins of rectal metastases were the breast (42 patients), stomach (38 patients), and prostate (16 patients). Rectal metastases occurred metachronously in the majority of patients (77%). The main treatment was surgical resection followed by chemotherapy [17].

The second patient with distant metastasis in the rectum was a 65-year-old man (patient no. 2), who had been diagnosed 8 years after radical prostatectomy for prostate cancer. Computer-assisted tomography revealed a large pelvic tumor encasing the rectum, which was histologically revealed to be a metastasis of the prostate carcinoma (Fig. 1). Cases of prostate carcinoma only very rarely metastasize to neighboring organs, such to the rectum as in this case [17]. Metastasis of prostate cancer to the rectum can occur in at least 3 possible ways: direct invasion through Denonvilliers' fascia, lymphogenic metastasis, or via

spreading of malignant cells into perirectal or rectal tissue as a result of a needle biopsy [19]. Despite radical resection of the metastasis with TME and a colonic pouch, the patient developed a recurrence in the bladder 6 months postoperatively and underwent cystectomy and resection of the colonic pouch. This was followed by adjuvant hormonal therapy involving abiraterone. The patient lived recurrence-free until the end of the follow-up period (48 months).

An 81-year-old female patient (patient no. 3) was diagnosed with **GIST** recurrence in the rectovaginal space with recurrent liver metastases. The initial diagnosis of GIST in the stomach had been made 35 years previously. Several recurrences had been resected over the years. According to the current literature, GISTs account for 20% of all soft tissue sarcomas. Most GISTs are located in the stomach (50-60%), followed by the small intestine (30-35%) and then the colon and rectum (5%). 40% of GISTs subsequently develop metastases. 10-20% of GISTs have already metastasized at the time they are diagnosed [21]. Imaging (tumor heterogeneity as a sign of malignancy), tumor size and tumor location (increased risk of metastasis if located in the intestine), and mitotic rate are all important for prognosis. Lymph node involvement is very rare, which is why lymphatic dissection is usually not necessary. Tumor rupture is recognized as an independent prognostic risk factor [20-22]. Decisive for GIST surgery is R0 resection while keeping the typically thin pseudocapsule intact. In advanced, metastatic GIST, imatinib can stabilize the course of the disease in over 80% of cases. Debulking surgery as the disease progresses is indicated particularly when alleviating symptoms is the goal. Radiotherapy can selectively contribute to the palliative effect [20-24]. In our case, the patient had another recurrence of a GIST tumor during therapy with imatinib (400 mg). The indication for surgical treatment was in order to provide symptom relief, as described in the literature. Due to the presence of a 2 cm supra-anal tumor with infiltration into the mesorectum and rectovaginal space, TME was performed with R0 resection. The administration of imanitib 400 was continued postoperatively. The liver metastases were treated using radiofrequency thermal ablation (RFTA). There was no indication for radiotherapy in the R0 resection.

In one male patient, adenocarcinoma arose from **heterotopia of the pancreas** in the rectum. During a preventive colonoscopy, an induration in the anocutaneous line was discovered in a 63-year-old male patient (patient no. 4). The biopsy revealed only inflammation. Due to clinical suspicions of malignancy, transanal tumor resection was performed, during which the induration was found to be ductal adenocarcinoma in ectopic pancreatic tissue (Fig. 2). Oncological rectal extirpation was then performed, including the tip of the coccyx, followed by adjuvant chemotherapy with FOLFIRINOX. The patient lived recurrence-free until the end of the observation period (24 months). The first case report of ectopic pancreatic carcinoma located in the rectum dates from 2010 [25]. Cases of ectopy in the lower gastrointestinal tract are extremely rare and occur almost exclusively in the rectum. Ectopic pancreatic or gastric mucosal tissue is the most prevalent [26-29]. Portions of ectopic salivary gland or prostate tissue may also be found in the rectum and anal area. Very rarely, cases of ectopy can represent the starting point for tumors [30-34]. Such ectopy is rarely accompanied by symptoms or complications and does not usually cause any differential diagnostic problems. Yet depending on the location, size, and type of ectopic tissue, it is also possible for complications to arise. These complications include inflammation, ulcerations, bleeding,

pyloric gland adenoma due to ectopic gastric tissue in the rectum and anus, along with inflammation and various ectopic pancreatic tumors due to pancreatic ectopic tissue in the rectum and anus. Pancreatic ectopy can lead to pancreatic ductal carcinoma, acinar cell carcinoma, intraductal papillary mucinous neoplasia, or endocrine tumors. Bleeding has been described as a complication of hetero-ectopic prostate tissue in the rectum and anus [33, 35].

Another 59-year-old man (patient no. 5) underwent a colonoscopy with biopsies from a stenosing tumor 7 cm from the anus due to hematochezia and abdominal pain. A histological examination of the biopsies revealed dedifferentiated adenocarcinoma in the rectal wall. Since no origin of the carcinoma from the rectal or anal mucosa was able to be shown, carcinoma arising from ectopic tissue was also considered a possibility. Even during the final examination of the specimen – after oncological resection – the origin of the ectopic tissue could not be determined with certainty. From a pathological perspective, it was most likely ectopic pancreatic CA or unusual urothelial carcinoma.

Squamous cell carcinoma was diagnosed in 5 cases (Fig 3). These were a 59-year-old patient (patient no. 6) with SCC in the middle third of the rectum 10 cm from the anus, a 67-year-old patient (patient no. 7) with supra-anal SCC, a 62-year-old patient (Patient No. 8) with metastatic stenosing SCC of the middle and upper third of the rectum, a 53-year-old patient (Patient No. 9) with SCC of the lowest part of the rectum with intramural metastasis with an aspect of a perirectal giant cyst and a 38-year-old patient (patient no. 10) with SCC measuring 10 cm diameter assumed to be of the Douglas type with multiple tumor infiltrations into the rectum and rectosigmoid as well as into the uterus. None of the SCC were in contact with the anus or anal canal. In the literature, the occurrence of SCC of the gastrointestinal tract is most commonly described in the esophagus or anal canal. Previous studies reported an incidence of less than 1% in the rectum [36]. Due to its rarity, the etiology of SCC of the rectum remains unclear, although it is associated with chronic inflammation and prior radiotherapy [37-40]. In a population-based cohorts analyses (Comparison of outcomes between rectal squamous cell carcinoma and adenocarcinoma) shows that patients with SCC have a better prognosis compared to those with rectal adenocarcinoma [41].

The trend today is to treat SCC of the rectum – and also SCC of the anal canal – with definitive chemoradiotherapy (5-fluorouracil and mitomycin). In metastatic disease, there is no clearly validated regimen, and treatment should be based on anal canal squamous cell carcinoma recommendations [42-43]. Patients in stage T3-4 N0 or T1-4 N+ with isolated or few small metastases may be operated on following radiochemotherapy. Otherwise, international guidelines recommend cisplatin-based palliative chemotherapy during the metastatic stage, possibly in combination with radiation [44]. If the approach is curative, follow-up care should be the same as for anal canal carcinoma [42].

In three cases (patient nos. 6, 8, and 10), we performed oncological resection after neoadjuvant radiochemotherapy. In one patient (patient no. 9) with intramural metastatic SCC and a perirectal giant benign cyst, open TME along with the cyst had initially been performed. As in another case with transanal full-thickness resection of the tumor (patient

no. 7), adjuvant radiochemotherapy was performed here. In one female patient (no. 8), palliative chemotherapy was initiated after 2 surgical revisions due to recurrences. All other four patients (patient nos. 6, 7, 9, and 10) lived without recurrence until the end of the follow-up period (5-60 months).

The oldest patient in this case series was 85 years old at the time of his initial visit (patient no. 11). He presented with a presacral cyst measuring 15 x 9 x 15 cm that had been known for 2 years. After the cyst was excised, histology revealed **mucinous cystic adenocarcinoma** (Fig. 4). Mucinous adenocarcinoma is an extremely rare disease that develops insidiously and has no obvious symptoms in the early stages. The treatment strategy involves complete surgical resection and adjuvant chemotherapy. Spontaneous rupture or iatrogenic rupture can lead to serious consequences such as implanted metastases in the peritoneal cavity and abdominal wall [45]. Mucinous colorectal adenocarcinoma responds poorly to palliative or adjuvant chemotherapy. Microsatellite instabilities are frequently found, which is why the effectiveness of immunotherapy, possibly in combination with chemotherapy and/or radiation therapy, has been the subject of study [45-47]. Our patient's case involved incomplete tumor resection. Rectal extirpation was recommended due to sphincter infiltration, although the patient refused this. The patient also refused chemotherapy. Two years later, the tumor progressed. Local excision was once again performed. The patient refused the recommended radiochemotherapy. Eight months later, locally advanced recurrence led to abdominoperineal rectal extirpation. However, the patient died 2 days following the operation due to pulmonary decompensation.

Adenocarcinoma of the rectum was also present in a 51-year-old patient (patient no. 12) who had an endoscopically impassable tumor 6 cm from the anus (tumor stage: cT4, cN2, M1hep, CRM positive; G2). Given that there was a threat of ileus, an ileostomy was inserted. During neoadjuvant therapy, the perforation of the carcinoma occurred with the development of Fournier gangrene up to the left gluteal area (Fig. 5). This resulted in an emergency operation and later (2 days later) to palliative intersphincteric rectal resection to treat the inflammatory focus. Palliative chemotherapy was then initiated. The development of Fournier gangrene associated with rectal carcinoma is rare. Individual cases of Fournier gangrene due to perforation at the base of rectal carcinoma were first described in 2016 [48]. Here and in 2 other publications with a total of 3 patients, therapy consisted of insertion of a stoma, surgical debridement, and antibiotics administration [49-50].

Rare adenocarcinoma of the rectum was found in three cases. One 31-year-old pregnant patient (patient no. 13) was found to have multifocal hepatic lesions suspected of being malignant during sonography shortly before her delivery date. On the same night, secondary cesarean section was performed, which revealed rectal cancer with multiple hepatic metastases. **Adenocarcinoma in pregnancy** is a rarity (with an incidence of 1:13,000) [2]. Only a few hundred cases have been published. The diagnosis is usually made at a late stage because non-specific symptoms such as bleeding and changes in bowel movements are attributed to other proctological diseases [3, 4]. The type of therapy and the prognosis depend on the tumor stage. Decisions regarding therapy must be made on an individual and interdisciplinary basis in consultation with the pregnant woman. In general, the objective is

radical removal of the tumor as early as possible. While chemotherapy (after the first trimester) can also be administered during pregnancy, radiation cannot [51-52]. With our patient, palliative chemotherapy was initiated, leading to remission of the rectal carcinoma and normalization of tumor markers, but the liver metastases remained unresectable. One year after diagnosis, sigmoidostomy was performed due to the appearance of ileus symptoms, followed by deep anterior rectal resection with en-bloc removal of the uterus. Later on, lung and bone metastases materialized. The patient died 9 months later due to tumor progression.

In one 52-year-old patient (patient no. 14), a presacral mass 8 cm in size was initially found and, after further diagnostics, **synchronous colorectal carcinoma** (supra-anal rectal carcinoma with sphincter infiltration and a stenosing second-carcinoma in the rectosigmoid junction) without evidence of distant metastasis was also found (Fig. 6). After being discussed at the tumor conference, the tumor (metastasis) was removed presacrally, and oncological sigmoid resection was performed. The rectal cancer was left untouched, whereupon the patient was scheduled for restaging after neoadjuvant treatment of the rectal cancer followed by TME. The patient died of a pulmonary embolism 12 months later without any recurrences.

Synchronous colorectal cancer accounts for approximately 3.5% of all colon cancers. This type of carcinoma has different clinical, pathological, and molecular features compared to solitary colorectal carcinoma. Overall, the prognosis for synchronous colorectal cancer does not differ significantly from the prognosis for solitary colorectal cancer. However, a slightly better prognosis has occasionally been described in patients with synchronous colorectal cancer [53]. When a presacral mass is present, various tumor entities can be considered in the differential diagnosis. Approximately two thirds of such tumors are congenital tumors, of which two thirds are benign and one third are malignant. If the tumors are assessed histologically, an extremely wide spectrum of different entities can be seen. They include epidermoid cysts, dermoid cysts, enterogenic cysts, teratomas, neurogenic tumors (e.g., schwannomas), osseous tumors, and various other tumors such as metastatic carcinoma as in our case, or even liposarcomas, fibrosarcomas, and leiomyosarcomas [54-62]. Examining the surgical specimen did not reveal any genetic predisposition – no evidence of hereditary non-polyposis colorectal carcinoma (HNPCC).

As the benign cause of tumorous rectal stenosis with pancolitis, we observed an inflammatory mass of the rectal wall in a 40-year-old patient (patient no. 15) caused by a **graft-versus-host disease (GvHD)**, which had occurred 5 years following allogeneic peripheral blood stem cell transplantation. GvHD with pancolitis is a complication of allogeneic hematopoietic stem cell transplantation. Surgical treatment is only required extremely rarely, such as in cases of ileus or massive rectal bleeding. Systemic steroids are the first line of therapy for GvHD. Infliximab or oral budesonide is helpful in patients with steroid-refractory disease, particularly in those with gastrointestinal involvement [63]. In the literature, one case report described a child with pancolitis who had not responded to regular immunosuppressive medications. The child underwent subtotal colectomy because of untreatable rectal bleeding [64].

Our patient also suffered from severe diarrhea (20x/d) with intermittent fever and abdominal pain. Regarding the inflammation, a protective loop ileostomy was initially created. Subsequently, there was persistent severe therapy-refractory inflammation in the diverted remaining colon, which was unresponsive to anti-inflammatory therapy and led to recurrent septic flare-ups. A proctocolectomy one year later was unavoidable.

Conclusion:

Rare tumors of the rectum and perirectal tissue pose a significant diagnostic and therapeutic challenge. Due to the heterogeneity and rarity of such tumors, the data from the literature is often insufficient and there is a lack of clear treatment recommendations. In our patient population, the majority of patients (n=12; 80%) underwent rectal resection with curative intent. To achieve an R0 situation, multi-visceral resection was often unavoidable (n=5; 33%). The procedures were able to be performed with few complications in the majority of patients and led to recurrence-free survival until the end of the follow-up period in 9 patients (60%). This case collective is intended to contribute toward improving the data situation for rare tumors of the rectum and pararectal adipose tissue. Further studies are required to better assess the characteristics of these tumors and thus further optimize the outcome for afflicted patients.

Legends

Figure 1. Metachronous metastatic prostate carcinoma in the mesorectum (presacral metastasis with rectal wall involvement).
(A) Computed tomography of the abdomen showing a large presacral mass in the lesser pelvis (arrow) infiltrating the dorsal and left rectal wall, measuring 7.2 × 5 cm.
(B) Histology (haematoxylin–eosin, ×40): tumour-free rectal mucosa in the upper field; atypical glandular structures in the submucosa (arrow).
(C) Immunohistochemistry (PSA, ×400): cytoplasmic PSA expression in atypical glands (arrow), confirming metastatic prostate carcinoma.

Figure 2. Ectopic pancreatic carcinoma of the rectum without identifiable pancreatic primary.
(A) Surgical specimen following rectal extirpation with coccygeal tip; supra-anal tumour indicated (arrow).
(B) Histology: mucosal surface (white arrow) and adenocarcinoma cells (black arrow).

Figure 3. Squamous cell carcinoma of the rectum.
(A) Magnetic resonance imaging showing a 7 × 6 cm tumour at the anorectal junction with intramural metastasis and cystic perirectal appearance (arrow).
(B) Surgical specimen demonstrating multiple 1–2 cm tumour foci (arrows) located 10 cm from the anal verge.
(C) Histology (haematoxylin–eosin, ×200): islands of atypical squamous epithelium within scar tissue (arrow).
(D) Immunohistochemistry (cytokeratin 5/6, ×200): strong positivity in squamous carcinoma cells within fibrotic tissue (arrows).

Figure 4. Perianal cystic mucinous adenocarcinoma.
(A) Clinical appearance (lithotomy position): marked swelling and induration of the scrotum (black arrow) and perineum (white arrow).
(B) Computed tomography showing a well-defined, irregular, fluid-containing perirectal mass with bilateral scrotal involvement, approximately 16 cm in diameter (arrow).

Figure 5. Perforated rectal carcinoma with Fournier gangrene during neoadjuvant chemoradiotherapy.
(A) Magnetic resonance imaging prior to treatment showing stenosing rectal cancer (cT4 cN2).
(B) Computed tomography during therapy demonstrating suspected perforation with left gluteal soft tissue emphysema (arrow).
(C) Intraoperative findings: left gluteal swelling (arrow).
(D) Surgical specimen showing rectal tumour (arrow).
(E) Postoperative findings following extensive perianal resection (arrow).

Figure 6. Synchronous colorectal carcinoma (triple tumour: rectum, sigmoid colon, and presacral lymph node metastasis).
(A) Magnetic resonance imaging demonstrating an 8 cm presacral mass (arrow).
(B) Surgical specimen after resection of the presacral lymph node metastasis (8 cm).

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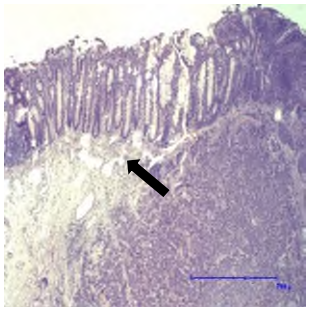
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Figure 1:

A.



B.



C.

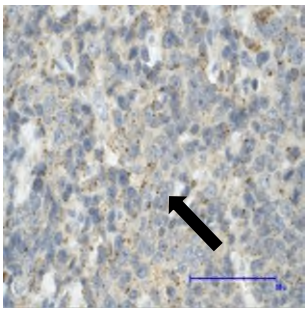
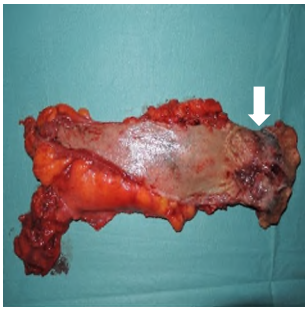
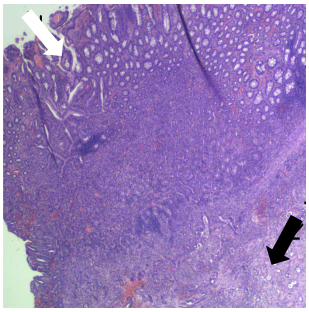


Figure 2:

A.



B.



C.

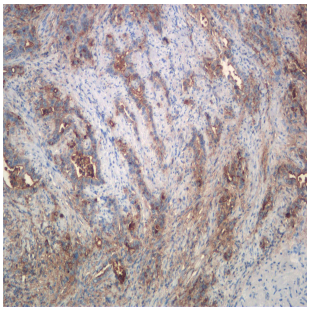
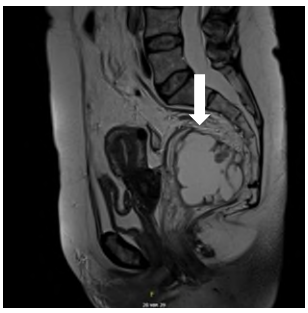
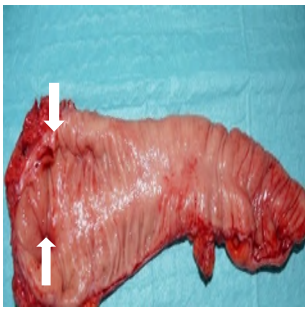


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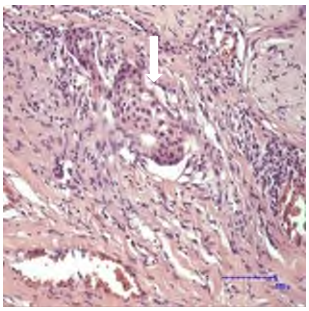
A.



B.



C.



D.

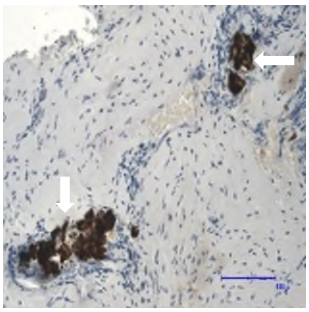


Figure 4:

A.

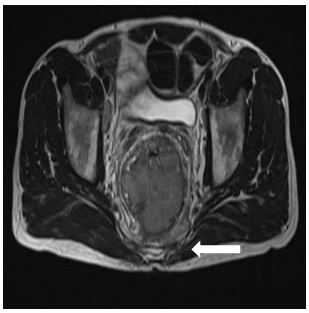


B.



Figure 5:

A.



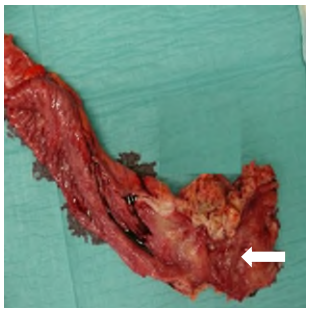
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D.

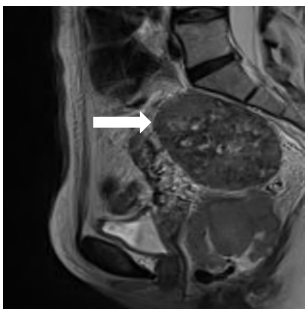


E.

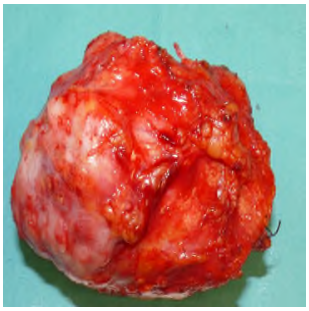


Figure 6:

A.



B.



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