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Case report

Isolated metastases to the common iliac and mesorectal lymph nodes suggesting an atypical metastatic route in upper rectal cancer: A case report

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ABSTRACT

Introduction: Most lymphatic vessels in the upper rectum run parallel to the superior rectal artery and up to the inferior mesenteric artery. Here, we report a rare case of upper rectal cancer with atypical lymphatic spread.
Presentation of case: A 39-year-old woman was diagnosed with upper rectal cancer and isolated lymph node (LN) metastases to the mesorectal and right common iliac LNs. The patient underwent laparoscopic low anterior resection with targeted dissection of the right common iliac LNs. The pathological diagnosis was T3N2M0, and the patient received postoperative adjuvant chemotherapy. One year later, local recurrence was found at the sacral promontory level, where the targeted lymphadenectomy had been performed previously. The recurrent tumor was surgically resected together with the attached presacral fascia. The patient subsequently received postoperative adjuvant chemotherapy, and there was no recurrence one year after the last surgery.
Discussion: Isolated metastases were observed in the right common iliac and mesorectal LNs. The locally recurrent tumor included lymphatic vessels running along the median sacral artery. No metastatic tumor was found in the internal iliac area at the time of the initial diagnosis nor during recurrence. Thus, this case suggests the presence of a rare metastatic route from the mesorectal LN to the common iliac LN via the median sacral lymphatics.
Conclusion: Lymphatic spread of rectal cancer may be predictable; however, rare patterns of LN metastasis can exist. The assessment of lymphatic flow is crucial for improving the oncological outcomes of rectal cancer surgery.

1. Introduction

Oncological efficacy in cancer surgery is achieved by obtaining adequate resection margins and eliminating metastatic lymphatic routes. The introduction and standardization of total mesorectal excision (TME) for rectal cancer has dramatically reduced the local recurrence rate and improved survival [1,2]. The development of neoadjuvant therapies, including radiotherapy (RT), chemoradiotherapy (CRT), and total neoadjuvant therapy (TNT), has contributed to enhanced local control; however, these treatment modalities are mostly indicative of lower rectal cancer [3,4]. Therefore, TME or tumor-specific mesorectal excision (TSME) is the mainstay of treatment for upper rectal cancer [5].

Previous anatomical studies have shown that most lymphatics in the upper rectum run parallel to the superior rectal artery and upward to the inferior mesenteric artery [6,7]. This concept has been widely accepted and rationalizes the oncological feasibility of TME or TSME for upper rectal cancer, in which sufficient mesorectal and inferior mesenteric trunk lymph nodes (LNs) are retrieved. However, surgeons may need to test their knowledge and practices when they experience unexpected cancer presentations.

Herein, we report a case of upper rectal cancer suggestive of atypical cancer spread with isolated LN metastases to the common iliac and mesorectal LNs. This study has been reported in line with the SCARE criteria [8].

2. Presentation of case

A 39-year-old woman visited our hospital with a chief complaint of bloody stools. Colonoscopy and contrast enemas revealed a type 2 ulcerated tumor in the upper rectum (Fig. 1a and b). Pathological diagnosis of the biopsied specimen confirmed the presence of

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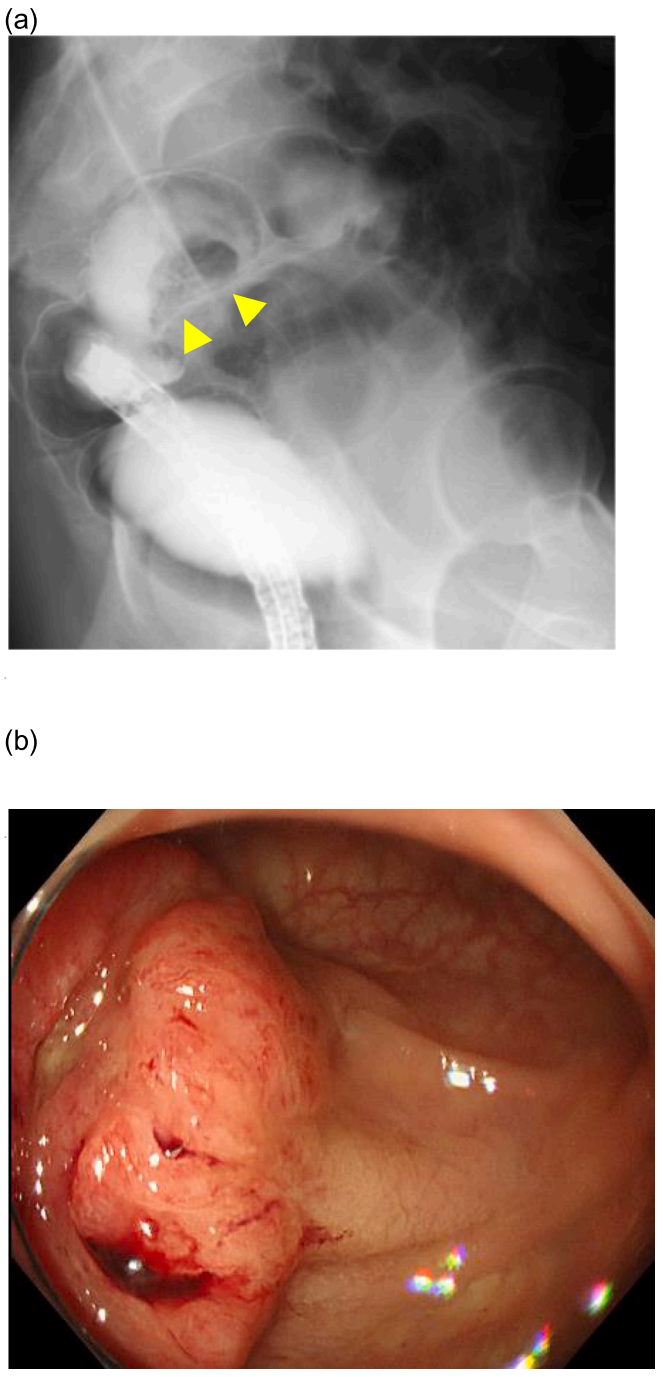

Fig. 1. Preoperative imaging studies.
(a) Contrast enema study revealed an irregular wall deformity in the upper rectum (arrowheads).
(b) Colonoscopy revealed an ulcerated lesion suggestive of type 2 advanced tumor in the upper rectum.
(c) PET-CT scan showed an increased FDG accumulation at the mesorectal (perirectal) lymph nodes (arrow).
(d) PET-CT scan showed an increased FDG accumulation at the right common iliac lymph nodes (arrow).

adenocarcinoma. A CT with IV contrast did not show any distant metastasis; however, the mesorectal (perirectal) LNs and right common iliac LNs were enlarged, suggesting LN metastasis. A subsequent PET-CT scan revealed increased FDG accumulation in the aforementioned area (Fig. 1c and d). The patient was diagnosed with stage IIIC rectal cancer (T3N2M0) according to the Union for International Cancer Control

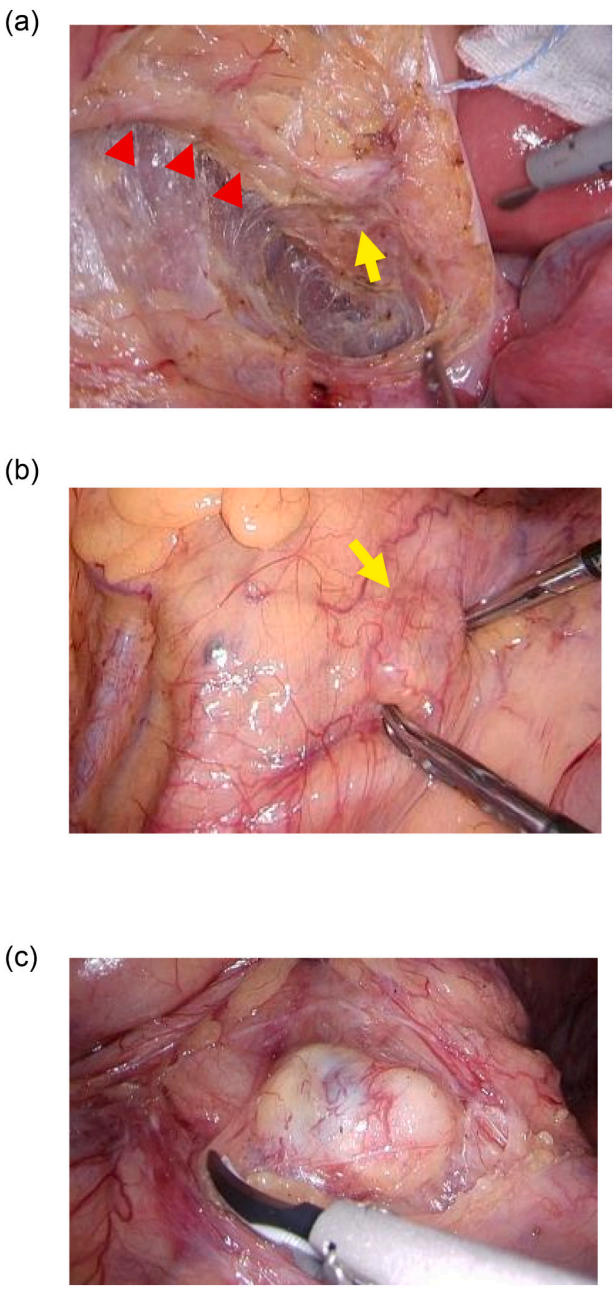

Fig. 2. Findings from the initial surgery.
(a) The mesorectal lymph nodes were exposed to the dorsal surface of the mesorectal fascia (arrow) and firmly attached to the hypogastric nerve (arrowheads).
(b) A solitary metastasis was found at the lymph node (arrow) ventrally to the right common iliac artery.
(c) The isolated common iliac LN was resected with the surrounding tissue.

(UICC) 8th edition classification [9] and scheduled for laparoscopic low anterior resection with TME or TSME, along with dissection of enlarged LNs around the common iliac vessels. According to the Japanese Society for Cancer of the Colon and Rectum (JSCCR) guidelines 2019 for the treatment of colorectal cancer, lateral LN dissection (LLND) is generally recommended for lower rectal cancer [10]. Therefore, considering the tumor location, we planned targeted LN dissection around the common iliac vessels.

During the surgery, the mesorectal LNs were exposed to the dorsal surface of the mesorectal fascia, extended to the presacral space, and firmly attached to the left hypogastric nerve (Fig. 2a). Therefore, en bloc

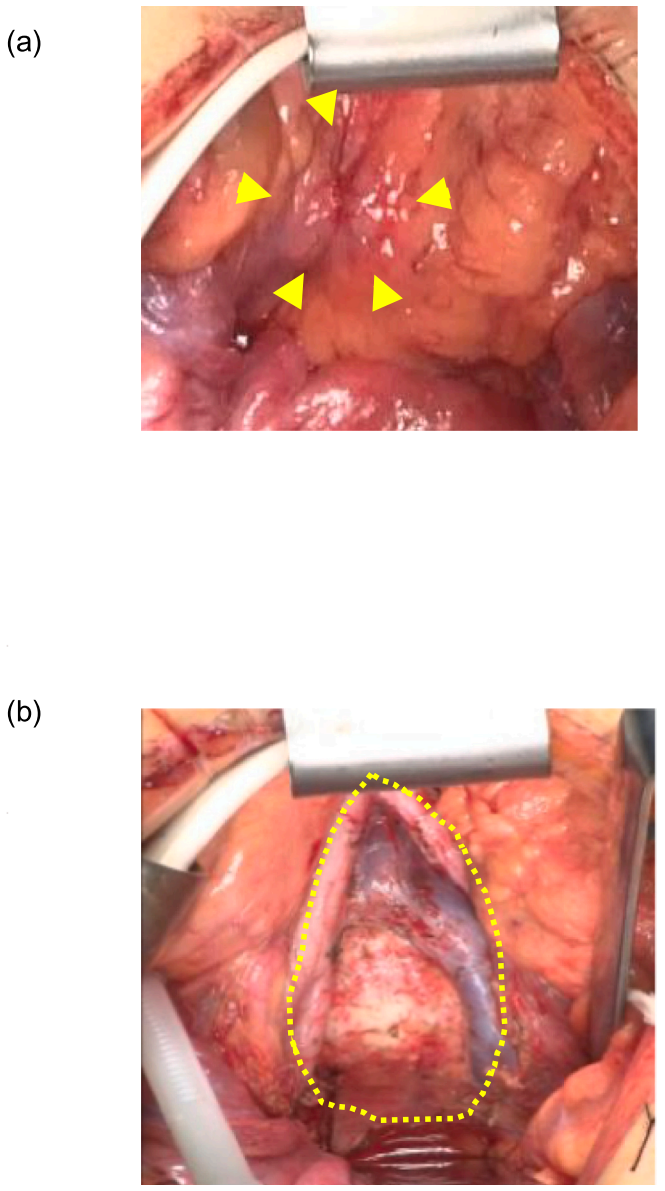

Fig. 3. Findings of the preoperative surgery.
(a) The recurrent tumor (arrowheads) is located at the level of the sacral promontory.
(b) Resected area (dotted line), including the recurrent tumor.

resection was performed along with the TSME. In addition, a solitary metastasis was found at the LN, located ventral to the right common iliac artery, consistent with the PET-CT findings (Fig. 2b). This LN was located 5 cm cranial to the previously dissected LN in the mesorectum without a direct connection (skip metastasis). The LN was mobile and resected separately from the surrounding tissue (Fig. 2c). After completing the TSME, colorectal anastomosis was performed using the double-stapling technique. Diverting stoma was not created. The patient's postoperative course was uneventful. The pathological diagnosis was T3N2bM0 stage IIIC according to the UICC 8th classification [9].

The patient received postoperative adjuvant chemotherapy with two cycles of the CAPOX regimen, followed by eight cycles of the capecitabine regimen. One year after surgery, PET-CT revealed a solitary mass with moderately increased FDG accumulation in the region where lymphadenectomy was performed, along with the right common iliac LN. Local recurrence was diagnosed and the patient underwent surgical resection of the recurrent lesion.

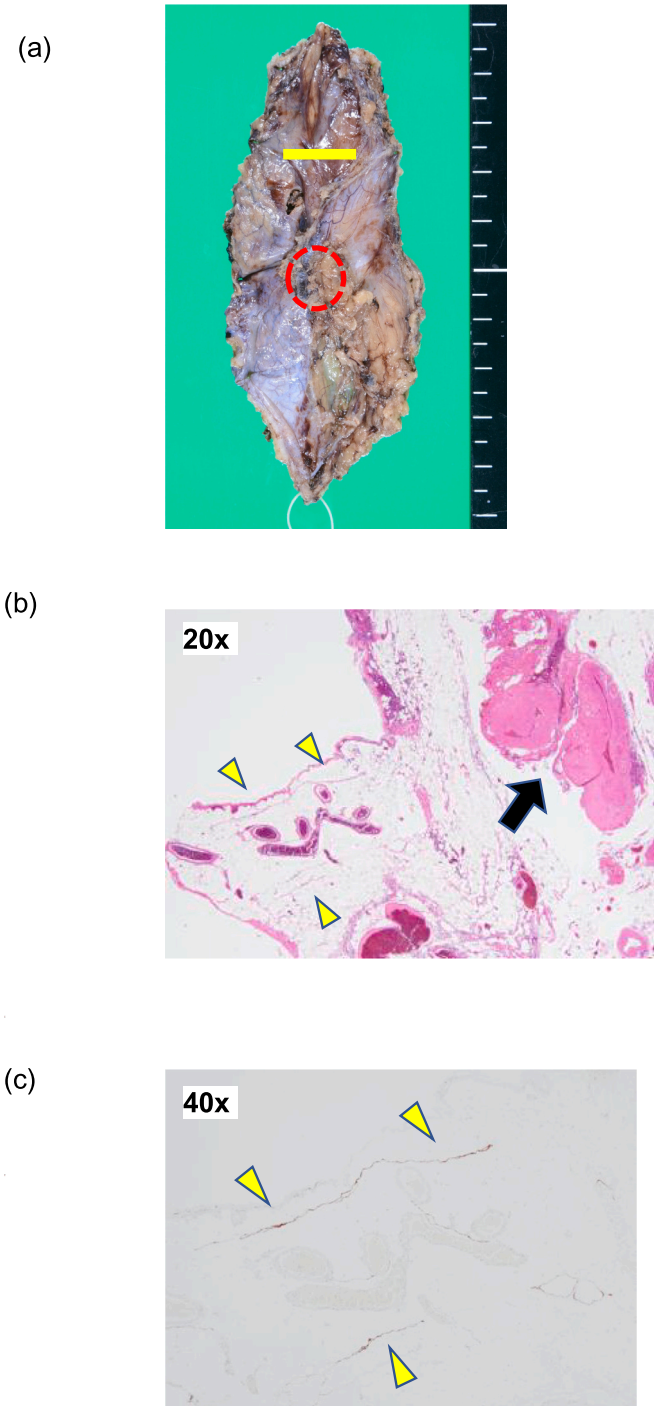

Fig. 4. Pathological examination of the resected specimen.
(a) The macroscopic view of the resected specimen.
(b) The microscopic view of the slice with the yellow line [in (a)] showed some collapsed lymphatic vessels (arrowheads) along with the middle sacral artery (arrow).
(c) The structure of lymphatic vessels was confirmed by the D2–40 staining.
(For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

Surgery was performed via open laparotomy. The recurrent tumor was located at the level of the sacral promontory and had partially invaded the left mesocolon (Fig. 3a). The tumor was resected together with the attached presacral fascia, 8 cm distal to the sacral promontory, preserving the left colic artery, right common iliac artery, and left colon (Fig. 3b). Since the anastomosis of the previous surgery was located 7 cm

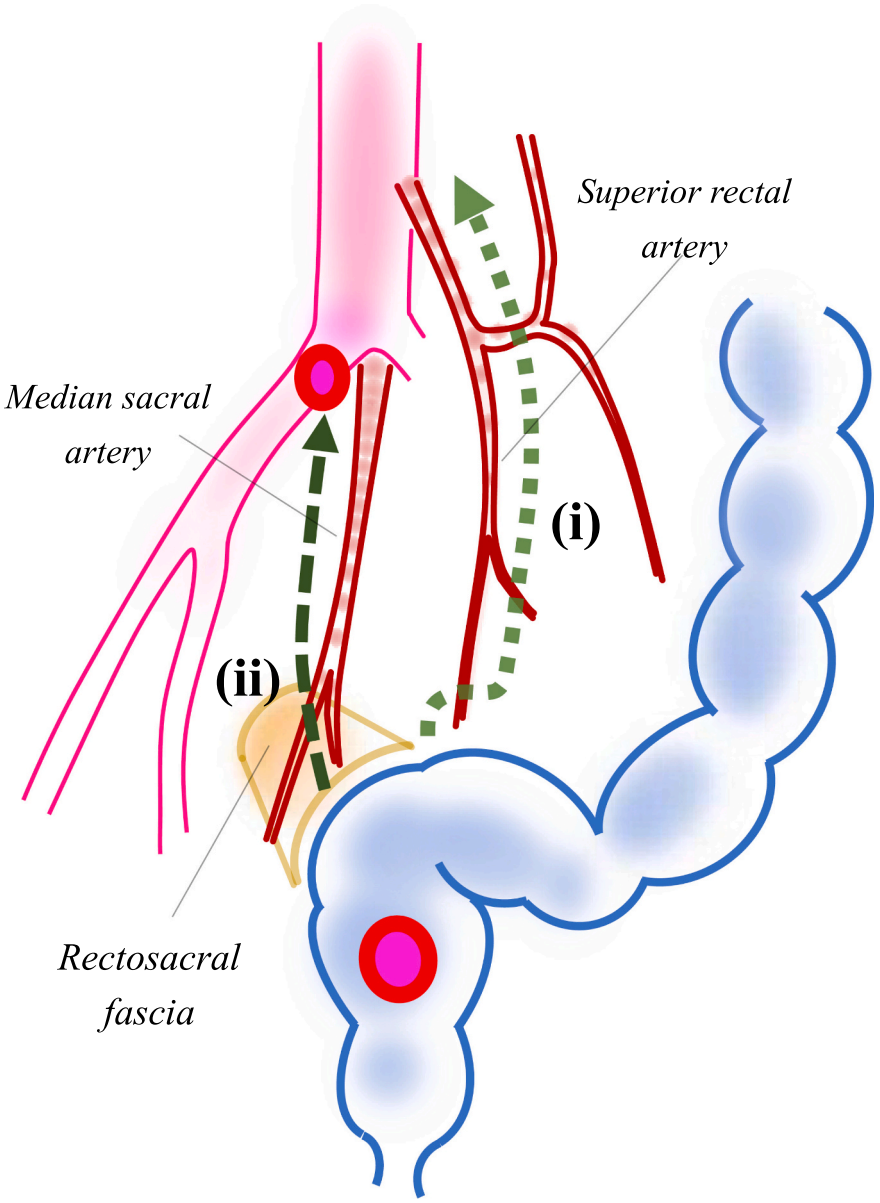

Fig. 5. Lymphatic spread of rectal cancer in this case.
(i) Typical LN metastasis route of upper rectal cancer along with the superior rectal artery.
(ii) Possible metastatic route to the common iliac LN via the median sacral lymphatics.

from the anal verge, it was isolated from the local recurrence site with an oncologically safe margin and was not included in the resected area. The postoperative course was uneventful. The pathological diagnosis of the resected specimen was compatible with local recurrence, and the resection margins were negative for cancer (Fig. 4a). At the upper 3rd level of the resected specimen, there were some collapsed lymphatic vessels running along the median sacral artery (Fig. 4b), as confirmed by D2–40 staining (Fig. 4c).

The patient subsequently underwent eight cycles of postoperative adjuvant chemotherapy with the mFOLFOX6 regimen, which was well tolerated. There was no recurrence one year after the last surgery.

3. Discussion

In rectal cancer, the following three routes have been considered as typical lymphatic spread: the upper route running alongside the superior rectal and the inferior mesenteric LNs, the middle route running across the lateral ligament to the iliac and obturator LNs, and the downward route to the inguinal LNs running either directly or indirectly through the internal and external iliac vessels [7,11,12]. Most lymphatic spread follows the upper route (within the mesorectum). In contrast, the middle and downward routes consist "extra-mesorectal" spread, which is characterized as the behavior of lower rectal cancer [7,13]. In this case of upper rectal cancer, isolated metastases presented at the right common iliac and mesorectal LNs and subsequently developed local recurrence at the same level in the presacral space. In addition, no metastatic tumor was found in the lateral LNs around the internal iliac area at the time of initial diagnosis or recurrence. Thus, this case suggests the presence of a rare but possible metastatic route from the mesorectal LN to the common iliac LN via the median sacral lymphatics (Fig. 5).

Within the rectosacral fascia, there are many branches of lateral sacral veins and sacral splanchnic nerves, whereas median sacral vessels are extremely rare. This suggests that few lymphatic vessels ascend directly to drain into the subaortic (aortic bifurcation) LN [14]. This may explain the rare incidence of metastasis to the median sacral LNs, which is as low as 1–2 % [15]. However, if the metastatic mesorectal LNs or the

tumor itself penetrates the outer line of the TME and attaches to or invades the hypogastric nerve or the pelvic nerve plexus, surgeons may need to consider possible metastatic lymphatics through the rectosacral fascia. The route could either be “lateral sacral to internal iliac route” or “median sacral to common iliac or subaortic route” in upper rectal cancer.

A similar case that showed atypical lymphatic spread of upper rectal cancer with isolated metastases to the mesorectal and median sacral LNs has been previously reported [16] and metachronous metastasis to the common iliac LN after curative resection of rectosigmoid cancer [17]. Moreover, a recent experimental study using intraoperative indocyanine green fluorescent imaging (ICG-FI) showed multiple lymphovascular connections between the mesorectum and presacral fascia as well as different patterns of lymphatic flow from the posterior rectal wall to the pelvic side wall and the presacral space up to the aortic bifurcation [12]. Taken together, “posterior dissection” with lymphadenectomy of the upward median sacral route may be indicated for selected cases, independently or in addition to standardized LLND, which retrieves the LNs around the internal iliac vessels.

Traditional anatomical investigations, functional analyses using lymphatic tracer injection, and isotope methods have contributed to an enhanced understanding of the lymphatic systems in the pelvis [6,7,13–15,18]. Currently, ICG-FI is used for real-time analysis of lymph node mapping [12,19]. Intraoperative assessment of tumor-specific lymphatic spread using fluorescence imaging may optimize the treatment strategies for locally advanced rectal cancer.

The drawback of this case report is that the patient developed local recurrence after the initial surgery, despite the completion of postoperative adjuvant chemotherapy. The reoperability of presacral and lateral recurrences for cure and prognosis is generally worse [20]. Furthermore, invasive local recurrences in the upper sacral and lateral areas may be associated with synchronous metastatic disease [20]. LN metastasis out of the main lymphatic drainage group may be considered non-regional or distant spread according to the American Joint Committee on Cancer guidelines [21]. Therefore, multidisciplinary consultations should have been conducted to discuss the possible indications for neoadjuvant therapy, including chemotherapy, RT, or CRT.

4. Conclusion

The lymphatic spread of rectal cancer may be predictable based on tumor location, whereas rare cases may present with atypical patterns of LN metastasis. Preoperative and intraoperative assessments of lymphatic flow are crucial for improved oncological outcomes in rectal cancer surgery.

Ethical approval

This case report has been approved by the Institutional Review Board of Tohoku Medical and Pharmaceutical University Hospital. Approved number: 2023-4-014.

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Writing-original draft: Tomoya Miura.

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All authors have read and approved the final manuscript for submission.

Guarantor

Shingo Tsujinaka, the corresponding author of this paper.

Research registration number

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Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

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Declaration of competing interest

All authors declare that there is no conflict of interest.

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