

Case Report

Successfully Resected Isolated Lateral Lymph Node Recurrence in a Patient with T1 Lower Rectal Cancer: Case Report and Literature Review

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Abstract

Lateral lymph node (LLN) metastasis in T1 rectal cancer has an incidence of less than 1%. However, its clinical features are largely uncharted. We report a case of LLN metastasis in T1 rectal cancer and review the relevant literature. A 56-year-old female underwent rectal resection for lower rectal cancer 2 years previously (pT1bN0M0). During follow-up, an elevated tumor marker CA19-9 was documented. Enhanced CT and MRI showed a round shape nodule 2 cm in size on the left side of pelvic wall. PET-CT showed high accumulation of FDG in the same lesion, leading to a diagnosis of isolated LLN recurrence. Because no other site of recurrence was detected, surgical resection of the LLN was performed. Microscopic findings were consistent with metastatic lymph node originating from the recent rectal cancer. Adjuvant chemotherapy for six months was given, and patient remains free of recurrent disease seven months after LLN resection. Although LLN recurrence after surgery for T1 rectal cancer is rare, post-surgical follow-up should not be omitted. When LLN metastasis is suspected on CT, MRI and/or PET-CT will be recommended. Surgical resection of LLN metastasis in patients with T1 rectal cancer may lead to favorable outcomes, when recurrence in other areas is not observed.

Keywords

case reports, rectal neoplasms, T1, lateral lymph node metastasis, lymphatic metastasis, literature review

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Introduction

Lateral lymph node (LLN) metastasis in lower rectal cancer has been observed in 16-23% of cases with a notably poor prognosis in metastatic instances[1]. Dissection of metastatic LLN does not improve long-term survival and only contributes to a reduction of the local recurrence rate[1]. Meanwhile, although surgeons have experienced limited cases that have been cured after resection of metastatic LLN, the features of patients with longer survival after resection of metastatic LLN is still uncertain.

In this report, we present a case of isolated lateral lymph node recurrence after resection of T1 lower rectal cancer. We also performed a literature review of similar cases. Interestingly, although an incidence of LLN metastasis in T1 rectal cancer is extremely small (<1%)[1], all seven cases reviewed showed favorable outcomes.

Case Report

A 54-year-old woman presented with a complaint of positive fecal occult blood test. Her past medical history in-

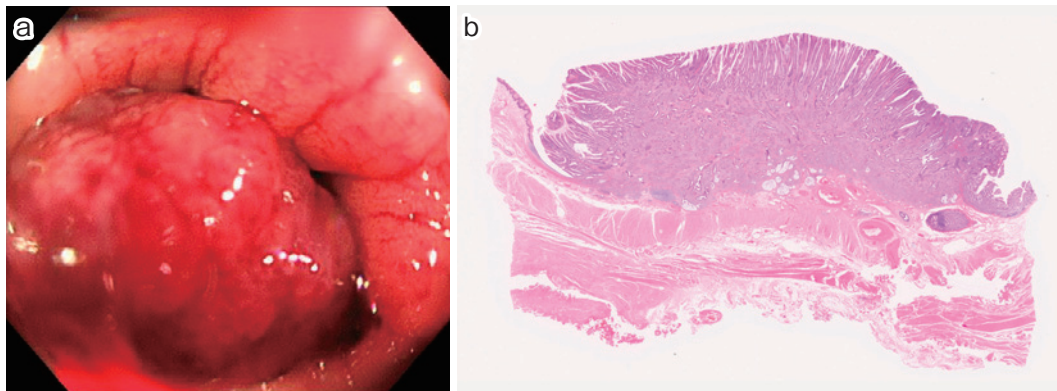


Figure 1.

a: A 0-Is lesion 2 cm diameter near the dentate line revealed by colonoscopy.

b: Histological examination of the resected primary rectal cancer showed a well differentiated adenocarcinoma invading the submucosal layer (8000 μ m, ly0,v1a).

cluded three years of treatment for hypertension. There was no family history of cancers including colorectum. Colonoscopy revealed a type 0-Is lesion in the lower rectum near the dentate line (Figure 1a), and biopsy showed tubular adenocarcinoma. Enhanced CT showed no lymph node nor distant metastasis. She was diagnosed with rectal cancer, and abdomino-perineal resection by open laparotomy method was performed. Pathology of the resected specimen revealed tubular adenocarcinoma (23×30 mm), T1b (SM 8000 μ m), Ly0, V1a, PM0, DM0, pRM0, pN0, and pStageI (Figure 1 b). No adjuvant chemotherapy was provided. Two years after rectal surgery, an elevated tumor marker CA19-9 (90 U/mL, normal range: <37 U/mL) was documented. The change of serum CA19-9 value during the perioperative period is presented in Figure 2. CEA value was within normal during the same period. Enhanced CT showed a round-shape nodule 2 cm in size on the left side of pelvic wall (Figure 3a). LLN recurrence was suspected, and she was referred to our hospital for further evaluation and treatment. PET-CT showed a high accumulation of 18 F-fluorodeoxyglucose (FDG) in the same lesion (Figure 3b). No other site of recurrence was detected. Contrast-enhanced MRI of the pelvis depicted a mass displaying a low signal on T2 (Figure 3c) and a slightly low signal on T1 (Figure 3d), leading to a diagnosis of isolated LLN recurrence. Uterine leiomyoma with malignant potential was also detected.

Because the enlarged LLN was isolated (No.263) and no other site of recurrence was detected, surgical resection of the lymph node by open laparotomy was performed. Concurrent total hysterectomy for uterine leiomyoma was also performed by gynecologists. Total blood loss during surgery was 610 g and duration was 341 minutes. Post-operative course was uneventful.

Macroscopic finding of the resected lymph node revealed a light yellowish solid mass measuring 31×27×18 mm (Fig-

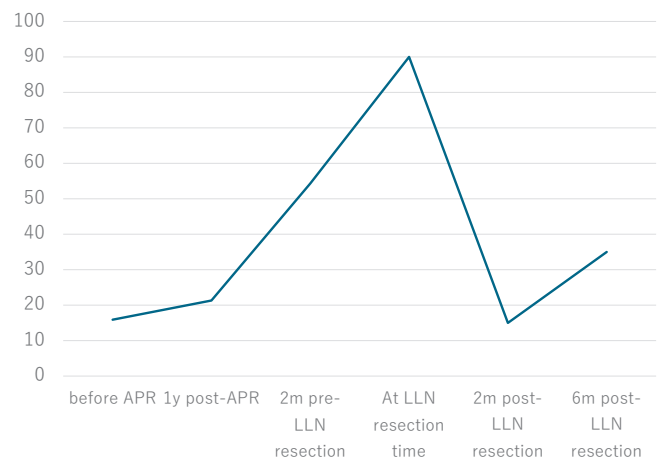


Figure 2. Serum CA19-9 values during a perioperative period are shown.

ure 4a). Microscopic findings were consistent with a metastatic lymph node originating from the recent rectal cancer (Figure 4b). No malignancy was observed in the uterus.

After six months of adjuvant chemotherapy with Capecitabine + Oxaliplatin (CapeOX), and the patient remains free of recurrent disease 7 months since LLN resection.

Discussion

Local recurrence including LLN metastasis in lower rectal cancer is a concern and often associated with a poor prognosis[2]. To reduce post-surgical local recurrence, neoadjuvant (chemo)radiotherapy with total mesorectal excision has been the standard of care in Western countries[3,4], while LLN dissection for advanced lower rectal cancer is generally performed in Japan. There is evidence that LLN dissection can lower the local recurrence rate[1]. Although LLN dis-

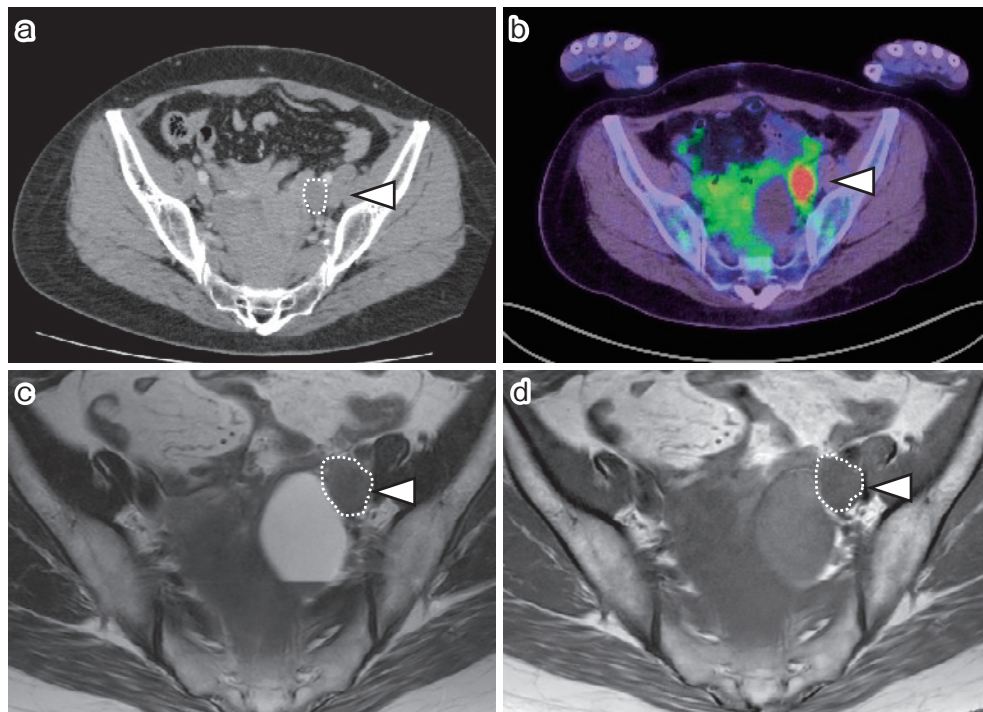


Figure 3.

a: Contrast enhanced CT showing round shape mass lesion on the left side of pelvic wall (arrow head).

b: PET-CT showing abnormal accumulation of FDG (arrow head).

c, d: MRI showing lymph node low on T2 and slightly low on T1 (arrow head).

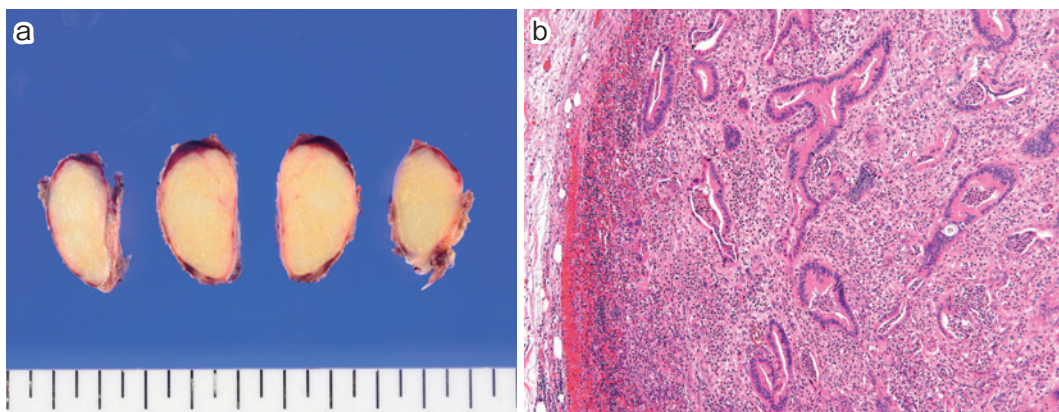


Figure 4.

a: Macroscopic finding showing a light yellowish solid mass.

b: Microscopic findings were consistent with metastatic lymph node originating from the recent rectal cancer (HE×100).

section does not contribute to improved survival, it is noteworthy that some patients showed longer survival by resection of positive LLN metastasis.

In the Japanese guidelines for rectal cancer treatment, LLN dissection is recommended for patients whose rectal tumors are located distal to the peritoneal reflection with the clinical diagnoses of >T3 or >N1 disease preoperatively[1].

Prophylactic LLN dissection is not routinely performed for T1 rectal cancer. The occurrence of LLN metastasis is extremely rare in T1 lower rectal cancer cases, accounting for only 0.9%[1].

We performed a systematic search of the Pub-Med database for English language literature published in and before September 2023 using the search terms 'rectal cancer', 'lat-

Table 1. Reported Cases of Lateral Lymph Node Metastases of T1 Rectal Cancer.

No.	Author	Publi- cation year	Age	Sex	Dif- feren- tiation	Dis- tance from anal verge	Size/shape of rectal tumor	Depth of invasion/Ly,v	Process of diagnosis	Time to LLNM m = months	LLNM size/ shape	Treatment	Mesorectal lymph nodes (positive/ total)	Lateral lymph nodes positive/total	Outcome (follow-up time, m=months)
1	Hara	2008	61	M	tub1	-	20 mm/0-Is	Not documented / Ly0,v0	Enlarged LLN on CT	51m	20 mm/ Round	openLAR → Open Right LLND	N0 (0/14)	1/10	alive (44m)
2	Sueda	2013	41	F	tub2	4 cm	23x20 mm/Not documented	Not documented / Ly0,v+	Enlarged LLN on CT	6m	26 mm/ Round	laparoscopic ISR → left LLND	N0 (0/24)	-	alive (12m)
3	Ogawa	2016	35	M	tub1	-	16x12 mm/0-IIa	3000 µm/ly1,v0	Enlarged LLN on MRI	simultaneous	4 mm/Round	laparoscopic LAR + Right LLND	-	-	alive (48m)
4	Tanishima	2017	56	M	tub2	12 cm	15x10 mm/0-Is	Head invasion/ly0,v0	Tumor marker elevation	6m	unmentioned/ Round	laparoscopic LAR → Open Right LLND	N0 (0/6)	3/15	alive (30m)
5	Zhang	2020	45	M	tub1, muc	10 cm	15 mm/Not documented	Not documented	Enlarged LLN on CT	Simultaneous	6.8 mm/ Round	ESD → Laparoscopic LAR + Left LLND	N0 (0/10)	1/11	alive (10m)
6	Liu	2022	36	F	tub2, muc	-	15 mm/0-Isp	Not documented	Enlarged LLN on CT	Simultaneous	4.8 mm/ Round	ESD → Laparoscopic LAR + Bilateral LLND	N0 (0/9)	2/22	alive (12m)
7	(present case)	2023	56	F	tub1	5 cm	23x30 mm/0-Is	8000 µm/ly0,v1a	Tumor marker elevation	27m	20 mm/ Round	Open APR → Open Left LLN resection	N0 (0/6)	1/1	alive (7m)

LLNM: Lateral Lymph Node Metastasis, LAR: Low Anterior Resection, LLN: Lateral Lymph Node, LLND: Lateral Lymph Node Dissection, EMR: Endoscopic Mucosal Resection, ESD: Endoscopic Submucosal Dissection, ISR: Inter Sphincteric Resection, APR: Abdomino Perineal Resection of rectum

eral lymph node metastasis' and 'T1', and only found 7 cases of lateral lymph node metastasis from T1 rectal cancers, including our case (Table 1)[5-10]. Mean age was 47.1y with 4 males and 3 females. Diagnoses of recurrence were from enlarged LLN on CT or MRI in 5 cases, and elevated tumor marker in 2 cases. In all cases, there was no regional (mesorectal) lymph node nor other metastases except for LLN observed at the time of rectal surgery. Disease free survival >6 months was obtained in all cases, including 3 cases >30 months, reinforcing the potential benefits of R0 resection of LLN metastasis in T1 rectal cancer. Three cases were with synchronous LLN metastasis, while 4 were metachronous (6-51 months), highlighting the importance of post-operative follow-up, even in Stage I cases. The Japanese guidelines for colorectal cancer treatment follow-up recommend tumor markers every three month, CT scan every six months, and colonoscopy every 1 year. The guidelines suggest that the interval between CT scans may be extended from 6 months to 1 year after 3 years follow-up for Stage I/II colorectal cancer. However, although rare, it should be understood that LLN recurrence may occur even after the third postoperative year.

Establishment of diagnostic criteria of LLN metastasis remains a pressing issue[1]. According to a retrospective study by the Japanese Colorectal Cancer Study Group, the diagnostic performance of MRI for metastatic LLN is better when the cutoff is 5 mm rather than 10 mm, but the study also showed the limitations of diagnostic performance based on lymph node diameter alone using conventional imaging method (sensitivity of 72.6%, and specificity of 54.7%)[11]. Also, there is a report that a 7 mm cut off value before neoadjuvant chemotherapy was able to predict LLN metastasis[12]. Diffusion-weighted MRI with conventional MRI for detecting LLN metastasis was reported to have sensitivity and specificity of 93% and 83% respectively[13], and those of FDG-PET was 83% and 95% respectively[14]. FDG-PET also has the advantage of excluding distant metastases. FDGPET and MRI is considered as a diagnostic option for LLN metastasis.

Therefore, as for the treatment strategy for LLN even in cT1 rectal cancer in our hospital, if LLN is more than 7 mm in short axis diameter, we will perform MRI and/or FDG/PET. If the lymph node shows abnormalities on MRI and/or FDG/PET, then we will decide to perform LLN dissection on the affected side. If both MRI and FDGPET are negative, follow-up will be recommended to the patient with informed consent. Otherwise, we will select LLN dissection on patient request.

In our case, although CT and MRI showed a 20 mm mass lesion in the pelvis, it was difficult to diagnose it as lymph node recurrence, because we believed LLN recurrence would be extremely rare in our case (pT1, pN0). There were also differential diagnoses including benign tumor (NET,

GIST, desmoid, schwannoma)[15,16]. Thus, positive PET-CT contributed proceeding to the surgery. We believe that PET-CT will be useful as a supportive diagnostic tool; however, routine PET-CT will not be necessary for general post-operative follow-up in patients with colorectal cancer.

Although LLN recurrence after surgery for T1 rectal cancer is rare, post-surgical follow-up should not be omitted. When LLN metastasis is suspected on CT, MRI and/or PET-CT are recommended. Surgical resection of LLN metastasis in patients with T1 rectal cancer may lead to favorable outcomes, when recurrence in other areas is not observed.

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Conflicts of Interest

There are no conflicts of interest.

Author Contributions

KR: conceived and designed the study. KK: revised the manuscript. All authors read and approved the final manuscript.

Approval by Institutional Review Board (IRB)

This study has received ethical approval from the Jichi Medical University Hospital Clinical Research Ethics Review Committee under reference number A21-064. Consent to publish patient data was received through the opt-out method.

Disclaimer

Toshiki Mimura is one of the Associate Editors of Journal of the Anus, Rectum and Colon and on the journal's Editorial Board. He was not involved in the editorial evaluation or decision to accept this article for publication at all.

References

1. Hashiguchi Y, Muro K, Saito Y, et al. Japanese Society for Cancer of the Colon and Rectum (JSCCR) guidelines 2019 for the treatment of colorectal cancer. *Int J Clin Oncol*. 2020; 25(1): 1-42.
2. Sugihara K, Kobayashi H, Kato T, et al. Indication and benefit of pelvic sidewall dissection for rectal cancer. *Dis Colon Rectum*. 2006; 49(11): 1663-72.
3. Monson JR, Weiser MR, Buie WD, et al. Practice parameters for the management of rectal cancer (revised). *Dis Colon Rectum*. 2013; 56(5): 535-50.
4. Glynne-Jones R, Wyrwicz L, Tiret E, et al. Rectal cancer: ESMO

- Clinical Practice Guidelines for diagnosis, treatment and follow-up. *Ann Oncol.* 2018; 29(Suppl 4): iv263.
5. Hara J, Yamamoto S, Fujita S, et al. A case of lateral pelvic lymph node recurrence after TME for submucosal rectal carcinoma successfully treated by lymph node dissection with en bloc resection of the internal iliac vessels. *Jpn J Clin Oncol.* 2008; 38(4): 305-7.
 6. Sueda T, Noura S, Ohue M, et al. Case of isolated lateral lymph node recurrence occurring after TME for T1 lower rectal cancer treated with lateral lymph node dissection: report of a case. *Surg Today.* 2013; 43(7): 809-13.
 7. Tanishima H, Kimura M, Tominaga T, et al. Lateral lymph node metastasis in a patient with T1 upper rectal cancer treated by lateral lymph node dissection: a case report and brief literature review. *Surg Case Rep.* 2017; 3(1): 93.
 8. Zhang X, He D, Ding Y, et al. Lateral lymph node dissection after endoscopic submucosal dissection for T1 rectal cancer: a case report. *ANZ J Surg.* 2020; 90(11): 2369-70.
 9. Ogawa S, Itabashi M, Hirose T, et al. Diagnosis of lateral pelvic lymph node metastasis of T1 lower rectal cancer using diffusion-weighted magnetic resonance imaging: A case report with lateral pelvic lymph node dissection of lower rectal cancer. *Mol Clin Oncol.* 2016; 4(5): 817-20.
 10. Liu XW, Zhou B, Wu XY, et al. T1 rectal mucinous adenocarcinoma with bilateral enlarged lateral lymph nodes and unilateral metastasis: A case report. *World J Clin Cases.* 2022; 10(33): 12404-9.
 11. Ogawa S, Hida J, Ike H, et al. Selection of Lymph Node-Positive Cases Based on Perirectal and Lateral Pelvic Lymph Nodes Using Magnetic Resonance Imaging: Study of the Japanese Society for Cancer of the Colon and Rectum. *Ann Surg Oncol.* 2016; 23(4): 1187-94.
 12. Sekido Y, Nishimura J, Fujino S, et al. Predicting lateral pelvic lymph node metastasis based on magnetic resonance imaging before and after neoadjuvant chemotherapy for patients with locally advanced lower rectal cancer. *Surg Today.* 2020; 50(3): 292-7.
 13. Mizukami Y, Ueda S, Mizumoto A, et al. Diffusion-weighted magnetic resonance imaging for detecting lymph node metastasis of rectal cancer. *World J Surg.* 2011; 35(4): 895-9.
 14. Rooney S, Meyer J, Afzal Z, et al. The Role of Preoperative Imaging in the Detection of Lateral Lymph Node Metastases in Rectal Cancer: A Systematic Review and Diagnostic Test Meta-analysis. *Dis Colon Rectum.* 2022; 65(12): 1436-46.
 15. Shin S, Maeng YI, Jung S, et al. A small, low-grade rectal neuroendocrine tumor with lateral pelvic lymph node metastasis: a case report. *Ann Coloproctol.* 2022; 38(4): 327-31.
 16. Watanabe Y, Baba H, Matsuzawa T, et al. [A Case of Pelvic Schwannoma, Mimicking Metastasis of Rectal Carcinoma]. *Gan To Kagaku Ryoho.* 2015; 42(12): 2262-4.

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