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World J Clin Cases 2018 October 26; 6(12): 483-576



REVIEW

- 483 Cancer and comorbidity: The role of leptin in breast cancer and associated pathologies
Ray A
- 493 One more chance of fistula healing in inflammatory bowel disease: Stem cell therapy
Turse EP, Dailey FE, Naseer M, Partyka EK, Tahan, V
- 501 Treat-to-target in Crohn's disease: Will transmural healing become a therapeutic endpoint?
Serban DE

ORIGINAL ARTICLE

Basic Study

- 514 CYP2C19 polymorphism has no influence on rabeprazole-based hybrid therapy for *Helicobacter pylori* eradication
Lin TJ, Lee HC, Lin CL, Wang CK, Chen KY, Wu DC

Retrospective Study

- 521 Declining diagnostic accuracy of non-invasive fibrosis tests is associated with elevated alanine aminotransferase in chronic hepatitis B
Wang L, Fan YX, Dou XG

CASE REPORT

- 531 Gemcitabine-induced haemolytic uremic syndrome, although infrequent, can it be prevented: A case report and review of literature
Cidon EU, Martinez PA, Hickish T
- 538 Colovesical fistula as the initial manifestation of advanced colon cancer: A case report and review of literature
Skierucha M, Barud W, Baraniak J, Krupski W
- 542 Robotic transoral vestibular parathyroidectomy: Two case reports and review of literature
Ozdenkaya Y, Ersavas C, Arslan NC
- 548 Atypical lipomatous tumor in the ligamentum teres of liver: A case report and review of the literature
Usuda D, Takeshima K, Sangen R, Nakamura K, Hayashi K, Okamura H, Kawai Y, Kasamaki Y, Inuma Y, Saito H, Kanda T, Urashima S

- 554 Computed tomography and magnetic resonance imaging findings of metastatic rectal linitis plastica from prostate cancer: A case report and review of literature
You JH, Song JS, Jang KY, Lee MR
- 559 Live birth after hysteroscopy performed inadvertently during early pregnancy: A case report and review of literature
Zhao CY, Ye F
- 564 Mesh migration into the sigmoid colon after inguinal hernia repair presenting as a colonic polyp: A case report and review of literature
Liu S, Zhou XX, Li L, Yu MS, Zhang H, Zhong WX, Ji F
- 570 *CNKSR2* mutation causes the X-linked epilepsy-aphasia syndrome: A case report and review of literature
Sun Y, Liu YD, Xu ZF, Kong QX, Wang YL

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Computed tomography and magnetic resonance imaging findings of metastatic rectal linitis plastica from prostate cancer: A case report and review of literature

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Abstract

Linitis plastica is a rare condition showing circumferentially infiltrating intramural anaplastic carcinoma in a hollow viscus, resulting in a tissue thickening of the involved organ as constricted, inelastic, and rigid. While most secondary rectal linitis plastica (RLP) is caused by metastasis from stomach, breast, gallbladder, or bladder cancer, we report an extremely rare and unique case of secondary RLP due to prostate cancer with computed tomography (CT) and magnetic resonance imaging (MRI) findings, including diffusion weighted imaging (DWI). A 78-year-old man presented with approximately a 2-mo history of constipation and without cancer history. On sigmoidoscopy, there was a luminal narrowing and thickening of rectum with mucosa being grossly normal in its appearance. On contrast-enhanced CT,

marked contrast enhancement with wall thickening of rectum was noted. On pelvic MRI, rectal wall thickening showed a target sign on both T2-weighted imaging and DWI. A diffuse infiltrative lesion was suspected in the prostate gland based on low signal intensity on T2-weighted imaging and restricted diffusion. A transanal full-thickness excisional biopsy revealed metastasis from a prostate adenocarcinoma invading the submucosa to the muscularis propria consistent with metastatic RLP. We would like to emphasize the CT and MRI findings of metastatic RLP due to prostate cancer.

Key words: Prostate cancer; Linitis plastica; Magnetic resonance imaging; Rectum; Metastasis

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Core tip: Secondary rectal linitis plastica (RLP) from prostate cancer is extremely rare. A target sign on T2-weighted imaging and diffusion weighted imaging is characteristic for RLP. The presence of elevated serum prostate specific antigen, T2 low signal intensity, and low apparent diffusion coefficient value lesion on prostate should raise the suspicion of secondary RLP from prostate adenocarcinoma. For confirmative diagnosis, full-thickness excisional biopsy is required due to its characteristic mucosal sparing.

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INTRODUCTION

Linitis plastica refers to a circumferential intramural tumor infiltration of a hollow viscus, generating a desmoplastic reaction that result in a rigid shrunken viscus with thickened walls^[1,2]. The stomach is the most commonly involved organ, although the small intestine, colon, and rectum also can be involved. This process can be either primary or secondary to metastasis, and shows characteristic mucosal sparing while involving only the submucosa and muscularis propria^[3]. Primary rectal linitis plastica (RLP), also known as signet ring cell carcinoma, is a very rare disease affecting less than 1% of colorectal cancer patients, while secondary RLP from stomach, breast, gallbladder, or bladder cancer occurs more frequently and has been reported previously^[4-8]. Due to the widespread use of pelvic magnetic resonance imaging (MRI) in rectal cancer evaluation, some characteristic findings have been suggested, such as a concentric ring pattern or target sign on T2-weighted imaging^[7,9].

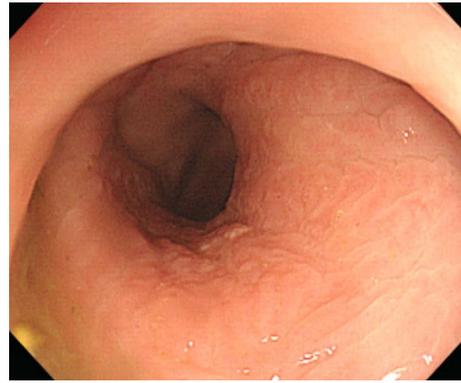


Figure 1 Sigmoidoscopic imaging showing narrowing of the rectal lumen with normal overlying mucosa.

Here we describe an extremely rare and unique case of metastatic RLP due to prostate cancer with findings of computed tomography (CT) and MRI including diffusion-weighted imaging (DWI). To the best of our knowledge, metastatic RLP as a first manifestation of prostate cancer with both CT and MRI findings has never been reported.

CASE REPORT

A 78-year-old man presented with a 2-mo history of constipation and thinning of stool caliber. He had a medical history of benign prostatic hyperplasia and hypertension. Laboratory findings were unremarkable, except the prostate-specific antigen (PSA) level was elevated to 409 ng/mL. Digital rectal examination revealed luminal narrowing of rectum with rigidity. Sigmoidoscopy revealed luminal narrowing of the mid to lower rectum, with grossly indurated, non-ulcerated mucosa (Figure 1). On contrast-enhanced CT, rectal wall thickening (mean thickness, 1.3 cm) with marked homogeneous enhancement and mild perirectal fat stranding was demonstrated (Figure 2A). Based on these observations, a submucosal spreading tumor was suspected, and pelvic MRI was performed for further evaluation. Coronal and sagittal T2-weighted imaging showed concentric thickening of the mid to lower rectum, about 6-7 cm in length, with stratification and perirectal fascial thickening. On axial T2-weighted imaging, the rectum showed three-layered wall thickening, with the middle layer showing isointensity and the inner and outer layers showing hypointensity (Figure 2B). On DWI, the lesion also revealed a concentric ring pattern or target sign, with the middle layer showing restricted diffusion while the inner and outer layers showed no restriction (Figure 2C). The prostate gland was enlarged and a diffuse infiltrative lesion was suspected showing low SI on both T1- and T2-weighted images with a low apparent diffusion coefficient (ADC) value (Figure 2D). There was also low SI on both T1- and T2-weighted images of the left seminal vesicle, and both external iliac lymph nodes were enlarged and round shaped. Positron emission

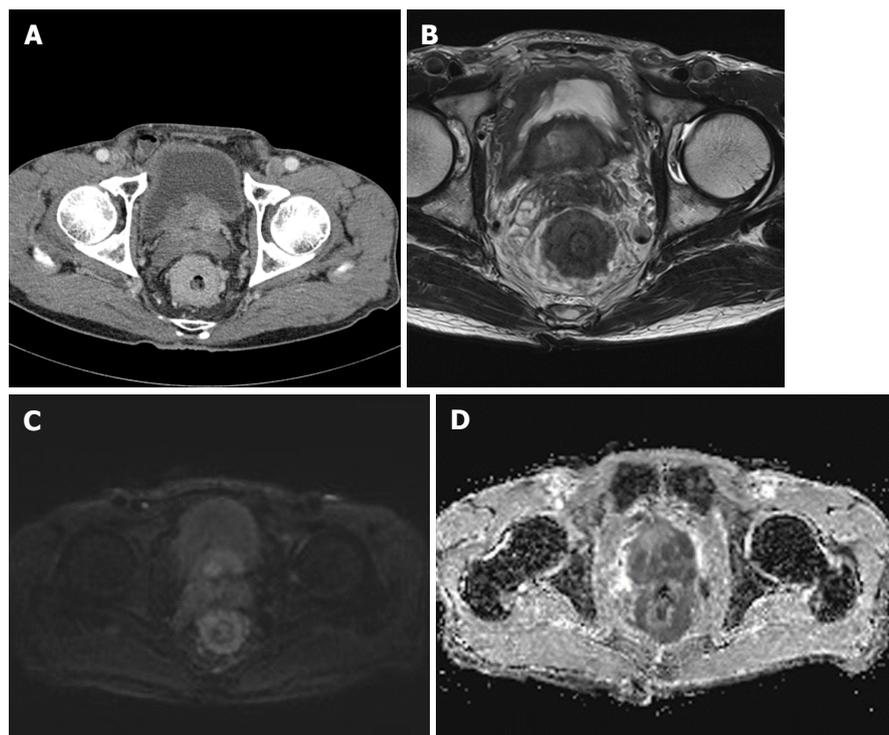


Figure 2 Contrast-enhanced computed tomography and pelvic magnetic resonance imaging of secondary rectal linitis plastica from prostate cancer. A: Contrast-enhanced CT shows circumferential rectal wall thickening with marked enhancement and minimal perirectal fat stranding; B: T2-weighted images; C: Diffusion-weighted images. On T2-weighted images and diffusion-weighted images, a target sign is present; D: Apparent diffusion coefficient map, the prostate shows infiltrative lesions with low ADC values, compatible with prostatic adenocarcinoma.

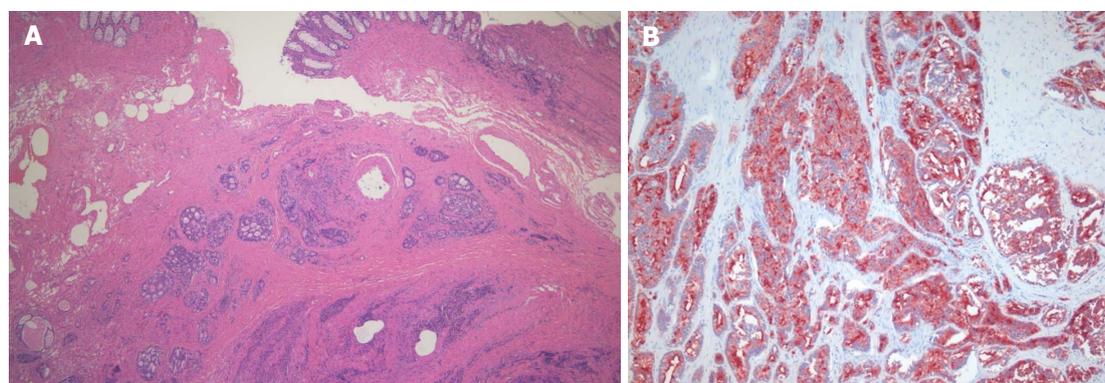


Figure 3 Histological images of metastatic prostate adenocarcinoma obtained from transanal full-thickness excisional biopsy. A: Hematoxylin and eosin (HE) staining showed a moderate to poorly differentiated adenocarcinoma characterized by tumor cells diffusely infiltrated into submucosa and muscularis propria, sparing the mucosal layer (original magnification $\times 100$); B: Immunohistochemical stains were performed and the tumor cells were positive for prostate specific antigen (PSA, original magnification $\times 100$).

tomography-computed tomography (PET/CT) revealed diffuse wall thickening of the mid to lower rectum without significant 2-deoxy-2- ^{18}F fluoro-D-glucose (FDG) uptake. There was no other pathologic site with abnormal FDG uptake. For both confirmative diagnosis and symptom relief, intraoperative transanal full-thickness excisional biopsy with loop sigmoid colostomy formation was performed. The pathologic examination revealed metastatic prostate adenocarcinoma invading the submucosa to muscularis propria with a positive PSA stain and negative for CDX2, consistent with metastatic RLP due to prostate cancer (Figure 3). Gleason score

of biopsy specimen was 9 (4 + 5). The patient underwent Luphere Depot (leuprolide acetate; a synthetic nonapeptide analog of naturally occurring gonadotropin-releasing hormone; used in the palliative treatment of advanced prostatic cancer) treatment and the PSA level decreased to normal without any adverse event. The patient has had no symptoms and his PSA levels have been normal for 2 years now.

DISCUSSION

Linitis plastica is characterized by diffuse proliferation of

connective tissue of a hollow organ wall by infiltrating tumor cells. This term was first introduced by Brinton W meaning the "leather bottle" shape of the stomach, and was later extended to other hollow organs to be constricted, inelastic, and rigid aspects of the wall^[10]. Primary RLP is very rare, while secondary RLP is more common, with some CT and MRI findings available for secondary RLP from breast and bladder cancer^[11-14]. To the best of our knowledge, there were only 2 case reports of secondary RLP from prostatic adenocarcinoma with only endoscopic ultrasonographic findings available^[5,15]. Therefore, this is the first case report to describe the CT and MRI findings of secondary RLP from prostatic adenocarcinoma.

Ha *et al*^[14] previously reported CT features of metastatic RLP in 22 patients. Primary cancer included 18 gastric cancer patients, 1 patient with bladder cancer (transitional cell carcinoma), 1 patient with ovarian cancer (serous cystadenocarcinoma), 1 patient with cervical cancer (squamous cell carcinoma), and 1 patient with colon cancer. Twenty patients (90%) had more than 5 cm in length of rectal involvement. In 19 patients (86%), rectal wall thickening (mean, 1.6 cm) extended to the lower rectum close to the level of the anal verge. In 6 patients (27%), the rectum showed three zones of layered wall thickening (target sign): hyperattenuated inner zone, hypoattenuated middle zone, and hyperattenuated outer zone. Of the 16 patients without the target sign, 3 showed strong contrast enhancement in the rectum. Our patient also showed marked rectal wall thickening with its length involving more than 5 cm, and showed marked contrast enhancement without a target sign on CT.

In the present case, T2-weighted imaging revealed doubled-layered concentric thickening of the rectal wall with an isointense middle layer and hypointense inner and outer layers representing a target sign. Based only on initial CT images, our first impression was infiltrative malignancy, such as lymphoma involving rectum, since smooth overlying mucosal thickening and concentric narrowing in the rectum was seen. However, there were only two lymph node enlargements at both external iliac chains, which is an unusual finding of lymphoma. Furthermore, other conditions such as chronic radiation colitis may contribute to rectal narrowing with circumferential wall thickening, but the patient did not have a history of pelvic malignancy or radiation therapy. On MRI, the involved rectum demonstrated a target sign on both T2-weighted imaging and DWI, which is considered as one of specific finding for diagnosing RLP^[12]. In addition, an infiltrative lesion with a low signal intensity on both T1- and T2-weighted imaging accompanied by heterogenous enhancement and low ADC values were revealed in the enlarged prostate gland. On PET/CT, there was no evidence of other sites with abnormal FDG uptake suggestive of metastasis or another primary cancer site. Based on these imaging findings and elevated serum PSA level, we suspected secondary RLP from prostatic adenocarcinoma.

In reviewing the literature, a characteristic pathologic finding of linitis plastica is an exuberant desmoplastic response that prominently elicits tumor cells and its product in submucosa and subserosa^[2,16,17]. It is difficult to diagnose on endoscopy in most cases because of its characteristic mucosal sparing. Therefore, cross sectional imaging findings may be helpful to guide early and precise diagnosis of RLP, whether primary or secondary. When there is a concentric rectal wall thickening with a layered appearance resembling a target sign, an experienced radiologist would check the patient's history to make a differential diagnosis such as inflammatory bowel disease (*e.g.*, ulcerative colitis, Crohn's disease), pelvic radiation (radiation proctitis), or primary malignancy (secondary RLP). If the patient has no such history, the possibility of either primary or secondary RLP should be raised, and workup for primary cancer detection is necessary.

In the present case, there was a target sign on MRI which also revealed a similar pathologic finding on full-thickness excisional biopsy, in which metastatic prostate adenocarcinoma invaded the submucosa and muscularis propria while sparing the mucosal layer. Although the direct comparison of whole rectum and MRI could not be made, we assume that this pathologic finding on the biopsy specimen was well correlated with T2-weighted imaging and DWI showing a concentric ring pattern or target sign, which represents a double-layered wall thickening. On DWI, diffusion restriction can be caused by highly cellular tissue (*e.g.*, tumor tissues), intracellular edema, increased viscosity, tortuous extracellular space (*e.g.*, fibrosis with or without granulation tissue) and increased densities associated with hydrophobic cellular membranes^[18]. We assume that in RLP, a tumor infiltrating the submucosa and muscularis propria associated with fibrosis and desmoplastic reaction would correlate with restricted diffusion in the middle zone, depicted as a target sign on DWI. The target sign could be a result of the normal zonal anatomy becoming more pronounced due to the invasion of an infiltrative tumor with fibrosis into the submucosa and on the longitudinal and circular layers of the proper muscle. In spite of substantial mural infiltration seen with RLP, the proper muscle layer is usually conserved, which may further explain the target sign^[14]. Further studies with whole rectal specimens to make precise correlations with MRI are needed.

In conclusion, when there is a long segment of circumferential rectal wall thickening with luminal narrowing, a target sign on T2-weighted imaging and DWI is suggestive of RLP. Therefore, knowledge of a patient's primary cancer history at other organs and the secondary findings in abdominal organs can suggest the possibility of secondary RLP rather than primary RLP.

ARTICLE HIGHLIGHTS

Case characteristics

A 78-year-old male patient was admitted because of a 2-mo history of constipation

and luminal narrowing of the rectum on sigmoidoscopy.

Clinical diagnosis

A submucosal spreading tumor of the rectum, such as lymphoma, was suspected.

Differential diagnosis

Chronic radiation colitis, inflammatory bowel disease.

Laboratory diagnosis

Elevated serum prostate specific antigen, suggesting prostate cancer.

Imaging diagnosis

Findings from pelvic magnetic resonance imaging led to a diagnosis of secondary linitis plastica of the rectum due to prostate cancer.

Pathological diagnosis

Metastatic rectal linitis plastica (RLP) due to prostate cancer.

Treatment

Loop sigmoid colostomy for immediate symptom relief and hormone therapy for prostate cancer.

Related reports

Metastatic RLP due to prostate is extremely rare, and this is the first case report to describe both computed tomography and magnetic resonance imaging findings.

Term explanation

There are no uncommon terms used in this manuscript.

Experiences and lessons

Although rare, careful evaluation with a high suspicion for other sites of malignancy, including the prostate, is needed when RLP is suspected on imaging studies.

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