Case Report

Linitis plastica of the rectum secondary to metastatic prostate cancer: A case report of a rare presentation and literature review

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Abstract

Linitis plastica is a rare tumor with poor prognosis. It is a circumferentially infiltrating intramural tumor which can result in rigid, nondistensible thickening of the affected organ. It most commonly affects the stomach, followed by the rectum, and can be due to primary or secondary cancer. Secondary rectal linitis plastica (RLP) caused by metastatic cancers has been reported from the stomach, breast, gallbladder, urinary bladder, and very rarely, the prostate, with only <5 reported cases in the literature. We report the case of a 66-year-old man who presented with altered bowel habit and loss of weight, with elevated prostate-specific antigen of 180.6 ng/mL. Sigmoidoscopy showed thickened rectal mucosa, and biopsy was negative for malignancy. Magnetic resonance imaging showed circumferential wall thickening, "target sign" appearance suggestive of RLP, PIRADS 5 lesion with extraprostatic extension, infiltrating bilateral seminal vesicles, and right neurovascular bundle. Repeat colonoscopy was performed under anesthesia, and deeper biopsy revealed poorly differentiated metastatic prostate adenocarcinoma. This case report highlights the atypical presentation of metastatic prostate cancer secondary to RLP, the rarity of this condition, and emphasizes the importance of deeper biopsy in RLP due to disease involvement predominantly in the submucosa and muscularis propria layers.

Keywords: Linitis plastica, metastatic, prostate cancer, rectum, secondary

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INTRODUCTION

Linitis plastica is a circumferential intramural tumor infiltration of a hollow viscus that causes desmoplastic reaction resulting in a rigid, constricted viscus with thickened walls. This condition was first reported by Brinton in 1859 and Borrmann in 1926, and it can affect the entire digestive tract either as primary or secondary cancer. The stomach was found to be the most commonly affected organ, accounting for about 68.3% of cases, whereas the rectum was the second most common (11.7% of cases).[1] Primary rectal linitis plastica (RLP) is very uncommon, with a reported incidence of <1% of all colorectal

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malignancies.[2,3] Secondary RLP, on the other hand, is more common, and the location of primary tumors has been reported from the stomach, breast, gallbladder, urinary bladder, and very rarely, the prostate. We herein report an atypical case of a 66-year-old man who presented with altered bowel habit due to severe anorectal stenosis caused by RLP which turned out to be metastatic prostate cancer.

CASE REPORT

A 66-year-old man with no significant medical history presented with constipation, which required daily digital rectal evacuation, tenesmus, fecal incontinence, and loss of weight (10 kg) over the duration of 3 months. He denied lower urinary tract symptoms or per rectal bleeding. He was an ex-chronic smoker, having stopped few years ago, and he had no family history of malignancy. Severe rectal stenosis with hard nodular mass (1 cm from the anal verge) was found on digital rectal examination. Prostate examination was not possible due to the low anal stricture which did not admit an examining finger. Sigmoidoscopy, performed using a small-caliber endoscope, noted thickened mucosa from anus to proximal rectum. Rectal mucosa biopsy reported infective colitis. Blood investigation revealed significantly elevated prostate-specific antigen (PSA) level (180.6 ng/mL) with normal levels of other tumor markers including carcinoembryonic antigen (1.1 ng/mL), CA19-9 (12.2 U/mL), and alpha-fetoprotein (3.67 IU/mL). Inflammatory markers were normal (C-reactive protein = 1 mg/L and erythrocyte sedimentation rate = 22 mm/h).

Contrasted magnetic resonance imaging (MRI) of the pelvis revealed long-segment circumferential wall thickening, involving all tissue layers of the rectum, of up to 20 mm in maximum thickness, extending from the mid-rectum to the mid-upper anal canal, beginning approximately 1.4 cm from the anal verge, and measuring about 9 cm in length. This caused significant luminal narrowing of the rectum and anal canal, resulting in mild dilatation of the proximal rectum. The T2-weighted images showed thickened hyperintense mucosa, thinned hypointense submucosa, and thickened isointense muscularis propria giving rise to a "target sign" appearance. On diffusion-weighted imaging (DWI), the mucosa and muscularis propria revealed restricted diffusion whereas the submucosa showed no restricted diffusion [Figure 1]. There was absence of fat plane between the prostate and the anterior rectum with tumor infiltration into the mesorectal fat, mesorectal fascia, and anterior peritoneal reflection. Post-gadolinium images showed marked contrast enhancement of all three layers of the rectum, anal canal, bilateral seminal vesicles, and prostate, along with multiple enhancing bilateral pelvic, internal iliac, external iliac, and bilateral inguinal lymph nodes [Figure 2]. Based on the elevated PSA and radiological findings, a provisional diagnosis of prostate cancer-causing secondary RLP was made. Using the tumor-node-metastasis staging, we staged this patient as T4 N1 M1.

The diseased lumen was too narrow for the insertion of a transrectal ultrasound probe for the purpose of either a classical transrectal or transperineal prostate biopsy to confirm the diagnosis of prostate cancer. Alternative methods to obtain a prostate biopsy were discussed in a multidisciplinary team meeting, including a transperineal prostate biopsy guided by transcutaneous ultrasound imaging of the perineum using a curvilinear or linear probe. A decision was made to attempt repeat biopsy of deeper layers of the rectum through colonoscopy under anesthesia. Colonoscopy showed a circumferentially thickened, hard mass with contact bleeding extending along the length of 2–10 cm from the anal verge [Figure 3]. The second biopsy showed poorly differentiated adenocarcinoma. Immunohistochemistry studies suggested metastatic prostate adenocarcinoma (positive PSA with negative CK7 and CK20) [Figure 4].

Prostate-specific membrane antigen ligand positron emission tomography showed uptake at prostate and rectal and anal walls with distant metastases at pelvic, cervical, and supraclavicular lymph nodes. Treatment options were discussed with the patient, which included palliative resection of the prostate, seminal vesicles, and anorectal canal, palliative chemotherapy, and androgen deprivation therapy (ADT). He refused major surgery which would

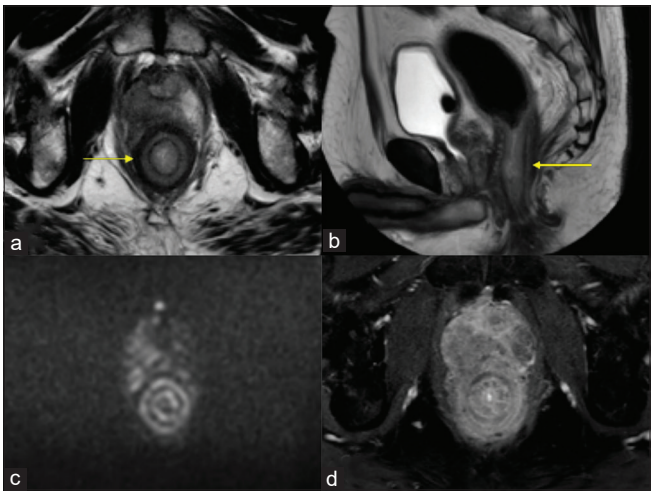

Figure 1: Magnetic resonance imaging pelvis (a) "target sign" appearance (arrowed) of rectal linitis plastica; (b) long anorectal thickening (arrowed) due to rectal linitis plastica causing obstruction and proximal mild rectal dilatation; (c) restricted diffusion of the mucosa and muscularis propria while no restriction on submucosa on diffusion-weighted imaging; (d) T1 axial fat suppression images of the rectum post-gadolinium

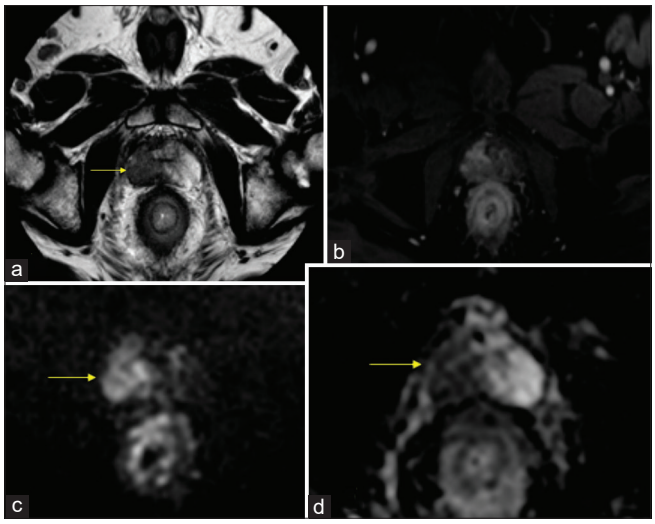

Figure 2: multiparametric magnetic resonance imaging prostate (a) PIRADS 5 lesion at right peripheral and transition zones (arrowed) with extraprostatic extension and local infiltration to bilateral seminal vesicles, right neurovascular bundle; (b) dynamic contrast enhancement post-gadolinium; (c) lesion markedly hyperintense on high b-value on diffusion-weighted imaging; (d) focal markedly hypointense lesion on apparent diffusion coefficient

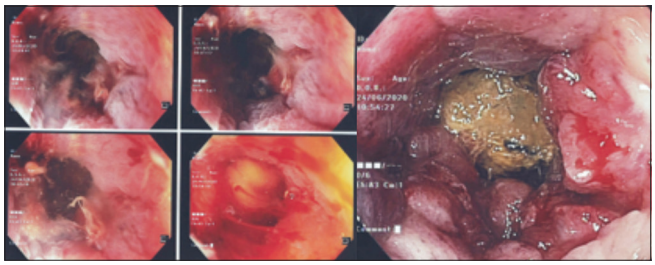

Figure 3: Colonoscopy showed circumferential thickening hard mass with contact bleeding extending along 2–10 cm from anal verge

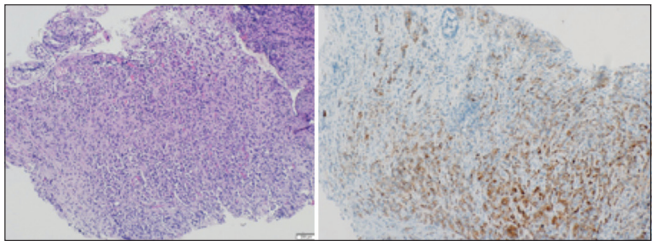

Figure 4: (Left figure): fragment of colonic tissue with infiltration of malignant cells in single and vague glandular formation. The malignant cells are enlarged, hyperchromatic with conspicuous nucleoli. (Right figure): the malignant cells are faily diffusely positive for prostate-specific-antigen

leave him with a permanent colostomy and upfront chemotherapy. He decided to receive ADT as the sole therapy. Serum PSA 3 months after starting treatment was 5.33 ng/mL, and his bowel habit improved significantly.

DISCUSSION

Prostate cancer is the third most common cancer among Malaysian men, and 53.2% of cases presented late with Stage IV disease.[4] These patients commonly present with lower urinary tract symptoms, urinary retention, gross hematuria, lower back pain, or neurological symptoms associated with spinal cord compression caused by pathological spine fracture. Very rarely do patients present with altered bowel habit due to bowel infiltration of the cancer, causing secondary linitis plastica, as the Denonvilliers' fascia usually prevents the posterior extension of prostate cancer. This mass could be mistaken as a rectal carcinoma and disastrously result in an inappropriate abdominoperineal resection.[5] To the best of our knowledge, there are only <5 reported cases of RLP secondary to prostate cancer.

It is a rare tumor with poor prognosis which can affect any part of the gastrointestinal system: stomach (68.3%), small bowel (5%), colon (10%), and anorectal (13.4%).[1] These tumors can either be a primary malignancy or secondary to metastasis or local infiltration from other sites such as breast, stomach, gallbladder, or urinary bladder.[3] There are only a few cases of secondary tumor from urological cancers reported in English literature – urinary bladder[6,7] and prostate.[3,8]

Obtaining a tissue biopsy to determine the primary tumor was challenging in our case. Endoscopic mucosal biopsy often fails to demonstrate the presence of malignancy because the disease usually affects the submucosa and muscularis propria layers,[8,9] as shown in the first initial biopsy of our patient, which was reported as infective colitis. In this case, prostate was deemed the most likely primary site based on the markedly elevated PSA level and suggestive MRI images. However, we could not insert the transrectal probe through the stenosed rectum, hence the conventional routes of prostate biopsy were impossible. We reviewed similar reported cases in the literature and their methods of obtaining tissue biopsy. You *et al*. performed transanal full-thickness excisional biopsy[3] and Bhutani performed endoscopic ultrasound (EUS)-guided fine-needle aspiration (FNA) of the deeper rectal wall which revealed metastatic prostate adenocarcinoma.[8] Initially, we had contemplated performing a transperineal prostate biopsy, guided by transcutaneous ultrasound imaging of the perineum using the curvilinear or liner probe. This idea was abandoned due to lack of experience of the surgeons and radiologists alike. After discussion with the colorectal surgeon, we decided to repeat colonoscopy under anesthesia to perform deeper biopsy which showed poorly differentiated adenocarcinoma.

Computed tomography (CT) or MRI scans are commonly used to assess the tumor. CT scan usually reveals diffuse,

circumferential wall thickening and delayed homogeneous enhancement associated with lack of distension, caused by the loss of bowel wall flexibility due to tumor infiltration.[1] On the other hand, concentric ring pattern perpendicular to the rectum, or "target sign" appearance, and significant rectal wall thickening are commonly observed on T2-weighted and T1-weighted MRI images. The ring pattern or "target sign" is thought to be caused by exaggeration of the normal zonal anatomy due to interposition of infiltrative tumor and fibrosis in the submucosa and around the circular and longitudinal layers of muscularis propria.[2,3] EUS has been considered useful in the diagnosis of RLP, where it is characterized by circumferential thickening of the rectal wall (with mean thickness of 13 mm) predominantly affecting the submucosa or muscular propria layers and associated with obliteration of the five-layer echo architecture of the rectal wall. However, it cannot differentiate between primary disease and secondary RLP.[8,9]

In conclusion, making a diagnosis of RLP is often difficult due to the nonspecific endoscopic findings and frequent negative mucosal biopsies due to deeper layer involvement. Imaging features suggestive of RLP on CT or MRI can aid the diagnosis, and these include circumferential wall thinking and the distinctive "target sign" seen on T2 images and DWI. Deeper endoscopic biopsy or EUS-guided FNA may successfully obtain tissue samples required for the pathological diagnosis of RLP.

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Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient (s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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