



# Endoscopic Submucosal Dissection of Rectal Cancer Close to the Dentate Line Accompanied by Mucosal Prolapse Syndrome

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### ABSTRACT

A 37-year-old man presented to our hospital for early rectal cancer accompanied by mucosal prolapse syndrome. Biopsy confirmed an adenocarcinoma, and endoscopic ultrasonography indicated proximity to the dentate line but no submucosal invasion. The tumor was removed en bloc via endoscopic submucosal dissection without complications, and its margin was free of tumor cells. The total procedure duration was 37 minutes, and the resected specimen measured  $23 \times 13$  mm. There was no recurrence during the 3-year observation period. Although close to the dentate line and accompanied by mucosal prolapse syndrome, a rectal cancer lesion was safely resected en bloc using endoscopic submucosal dissection.

### INTRODUCTION

Mucosal prolapse syndrome (MPS) is a rare and benign condition caused by occult or obvious mucosal prolapse. There are few reports of MPS accompanied by adenocarcinoma.<sup>1</sup> For these cases, the initial treatment is usually surgical; however, there have been no reports of the efficacy and safety of endoscopic submucosal dissection (ESD) of rectal cancer accompanied by MPS.

### CASE REPORT

A 37-year-old man was referred to our facility for the management of early rectal cancer. He had been diagnosed with MPS at another hospital after a biopsy revealed suspected adenocarcinoma with MPS. He reported no abnormal bowel habits. Colonoscopy in our hospital revealed an elevated lesion with redness, approximately 15 mm in diameter, extending close to the dentate line (Figure 1). Narrow-band imaging could not detect any surface pattern, as it was covered by a white coat (Figure 1). Biopsy specimens also indicated adenocarcinoma and fibromuscular obliteration (Figure 2). Endoscopic ultrasound (EUS) showed a hypoechoic mass in the second layer, while the third and fourth layers were intact (Figure 3). The lesion was diagnosed as early rectal cancer with MPS. ESD was selected for tumor resection.

The procedure was performed under conscious sedation with pethidine and midazolam. Diluted lidocaine with indigo carmine was used for a submucosal injection because of its location close to the dentate line. The procedure was completed in 37 minutes without complications. The tumor was removed en bloc, and its margin was free of tumor cells (Figure 4). Follow-up colonoscopy performed 3 years later showed no recurrence of rectal cancer and MPS.

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Figure 1. (A) Endoscopic view of the polypoid lesion approximately 15 mm in diameter. (B) Narrow-band imaging did not detect any surface pattern, as it was covered by a white coat. (C) Location of the lesion close to the dentate line.

#### DISCUSSION

This is a very rare case of rectal cancer accompanied by MPS, and it is the first report in which ESD was selected as the treatment for the combined conditions. MPS was first reported by Madigan and Morson as solitary rectal ulcer syndrome in 1969.<sup>2</sup> MPS is a benign disease caused by occult or obvious mucosal prolapse with fibromuscular obliteration that is diagnosed histologically. Patients with MPS usually have symptoms of abdominal discomfort or rectal bleeding, as well as abnormal bowel habits such as constipation or straining. Approximately 26% of patients report no symptoms or abnormal bowel habits.<sup>1</sup>

The endoscopic findings for MPS are categorized into ulcerative (55.1%), polypoid (24%), and flat (20.9) types.<sup>3</sup> Because of the various presentations, it is also important to consider the possibility of rectal cancer. Many reports regarding the distinction between MPS and rectal cancer have been published. However, only a few reports have described cases of MPS accompanied by rectal cancer. There has been speculation about the possibility of malignant transformation of MPS, but there have been no reports to support this hypothesis.<sup>1</sup> Therefore, the relationship between MPS and rectal cancer remains unclear.

Numerous reports have mentioned that erroneous histological findings lead to unnecessary surgical interventions. As MPS consists of irregular glands in some cases, MPS may erroneously be diagnosed as adenocarcinoma.<sup>3</sup> Several reports have indicated the efficacy of EUS or magnetic resonance imaging for distinguishing MPS from carcinoma. However, the use of these imaging methods does not decrease the incidence of misdiagnosis between MPS and rectal cancer.<sup>45</sup> In our case, tissue biopsy was performed twice in 2 hospitals, and both pathologists diagnosed the lesion as an adenocarcinoma. We used EUS not only for diagnosis, but also to determine whether the submucosa was intact; this meant that the risk of metastasis was negligible and thus ESD was indicated.

An optimal treatment for rectal cancer accompanied by MPS has not been established. Surgery, including transanal



**Figure 2.** Biopsy showed a well-differentiated tubular adenocarcinoma in situ, with fibromuscular obliteration in the lamina propria mucosa.



**Figure 3.** Endoscopic ultrasound showing a hypoechoic mass with no invasion of the submucosal layer (arrow).



Figure 4. The resected specimen was cut into 9 pieces for pathology; the red lines were indicative of adenocarcinoma.

endoscopic surgery, is the cornerstone of curative therapy for rectal adenocarcinoma. Kawaguti et al. reported on the effectiveness of ESD for early rectal cancer. They mentioned the possibility that ESD may be associated with a lower local recurrence rate than transanal endoscopic surgery, although no statistical significance was shown due to the small sample size and the retrospective nature of the study.<sup>6</sup> Meanwhile, there are several reports of ESD used for MPS because symptoms of bleeding or rectal prolapse are present, but they do not address any cases of adenocarcinoma.<sup>7</sup> Although the efficacy of using local lidocaine injection for lower rectal polyps has been reported, using ESD for lesions close to the dentate line is still considered a challenging procedure.<sup>8</sup> Moreover, elevated lesions usually make it difficult to determine whether the submucosal layer is intact. We performed ESD as an initial therapy for the following reasons: first, the EUS findings showed that the rectal cancer was in an early stage; second, the use and effectiveness of ESD for rectal cancer close to the dentate line has been reported; third, en bloc resection using ESD may be a more desirable and less invasive procedure compared to surgery. ESD was performed successfully without complications.

## DISCLOSURES

Author contributions: All authors contributed equally to the manuscript. H. Honda is the article guarantor.

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Informed consent was obtained for this case report.

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