

Debulking Operation, Radiation Therapy, and Sphincter-Sparing Local Wide Excision for The Lower Rectal Melanoma: A Case Report

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Case report

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Abstract

Background

Rectal melanoma is a rare disease that accounts for about 1% of rectal cancers. Abdominoperineal resection was the standard surgical intervention for local control. However, it can lead to complications and reduce the quality of life due to colostomy. Debulking surgery, radiotherapy (RT), and sphincter-sparing wide local excision (WLE) were performed on a patient with rectal melanoma.

Case presentation

A case of a 79-year-old woman with anal pain and bloody stool for 1 month was reported in this study. The digital examination of the rectum revealed a big polypoid mass over the lower rectum. Stage II rectal melanoma was diagnosed by colonoscopy, biopsy, magnetic resonance imaging, and positron emission tomography. The patient initially received debulking surgery to relieve the symptoms of active bleeding followed by radiotherapy for the residual tumor with partial response. Finally, a sphincter-sparing WLE was performed for the residual tumor to preserve the anal function. The postoperative course went smoothly. No local recurrence and anal symptoms were observed during the 2-years follow-up period.

Conclusions: Combining debulking surgery, RT, and WLE may be a viable alternative for rectal melanoma that cannot be completely resected at the beginning and accompanied by bleeding symptoms.

Background

Rectal melanoma is a rare disease that occurred in about 1% of rectal cancers [1] with a poor prognosis which is mainly implanted on or above the dentate line in women after 50 years old [2]. Common clinical symptoms are anal bleeding, anal pain, and changes in bowel habits. Therefore, it is initially often misdiagnosed as hemorrhoids or polyp. The tumor macroscopically presents as a polypoid mass in the lumen or thickening of the peripheral wall. On magnetic resonance imaging (MRI), rectal melanoma usually presented as an intraluminal polypoid mass with high signal intensity on T1-weighted images and low or mix-signal intensity on T2-weighted images with moderate to strong enhancement and diffusion limitation [3]. Due to its high sensitivity and specificity, positron emission tomography-computed tomography (PET-CT) was regarded as suitable for cancer staging and treatment response evaluation.

The histological markers, S-100 and HMB-45, are frequently identified microscopically [4]. A previous study revealed the poor prognosis that the median overall survival ranged from 8.0 to 18.6 months [5]. No strong evidence exists for diagnosis guidelines owing to rarely available data. The radical surgery with abdominoperineal resection (APR) was regarded as the first option owing to its better outcome in the early years [6–8]. However, it could lead to complications and reduce the quality of life due to colostomy. Sphincter-sparing wide local excision (WLE) has been gradually considered as an alternative option in recent years for better functional performance management and to decrease surgery morbidity than APR

[9, 10]. However, many disadvantages exist in clinical WLE use, including a tumor size that is too large to be initially completely resected and high local recurrence after surgery [7, 11–13]. Malignant melanoma was previously regarded as a radioresistant tumor. Since the late 1980s, more studies have indicated that WLE combined with adjuvant radiation therapy (RT) had been adopted to reduce the local recurrence in the recent 20 years [13–15]. Moreover, an adequate surgical approach may be considered in the current study to minimize surgical complications, achieve margin-free resection, reduce local recurrence, and maintain the quality of life of the patient. Thus, the case of an elderly with lower rectal melanoma was reported. This study aims to combine debulking operation, radiotherapy, and WLE as appropriate strategies to balance the patient's life functional quality and disease progression.

Case Presentation

A case of a 79-year-old woman who complained about perianal pain with bloody stool for 1 month was reported. The patient presented in good condition with neither weight loss nor previous medical history. The digital examination of the rectum revealed a big polypoid mass over the lower rectum. Moreover, a colonoscopy revealed a polypoid mass of about 6 cm in size and 5 cm above the anal verge (Fig. 1A). Tissue biopsy was performed with histopathology results showing malignant melanoma. The immunohistochemistry tumor profile was classified as Melan-A (+), S-100 (+), and ki-67 (+, 50%; Fig. 2). The abdominal MRI revealed a lobulated mixed-signal intensity mass at the rectum (5.5 × 4.8 cm) on the T2-weighted axial image (Fig. 1B). PET–CT revealed two intense FDG avidity over the rectum (standardized uptake value (SUV) = 16.1 at 5 o'clock and 11.3 at 9 o'clock) and one mild FDG avidity over the pre-sacral space (SUV_{max} = 4.6) suspicious nodal metastasis (Fig. 3). Rectal melanoma with regional lymph node metastasis, stage II [16], was complicated with diagnosed anal bleeding. Due to a multidisciplinary team discussion for the patient, the surgeon performed the first debulking operation surgery (Fig. 4) for symptom relief, combining with hypofractionated RT because the patient refused APR and immunotherapy. Follow-up abdominal CT revealed a striking shrinkage mass at the rectum with improved status compared with the previous image. The patient underwent secondary surgery with trans-anal WLE 3.5 months later for residue tumor resection after debulking operation. The patient was with clinical examination and image (CT scan or colonoscopy) for every 3 months follow-ups (Fig. 5). The patient is currently with 2-year relapse-free post-surgery.

Discussion And Conclusions

Rectal melanoma is generally an aggressive disease that has a poor overall 5-year survival rate, disease-free survival, and median survival period [6, 17–19]. More than half of the patients were found with regional lymph node metastasis at the time of diagnosis, and about 30% of the patients had metastasis to a distant location (e.g., lung, liver, bone, and brain) [20, 21]. The rectal melanoma surgical guideline is without consensus. Moreover, APR surgery had been regarded as the choice which performed a better margin-negative resection to control the lymphatic spread and local recurrence [22–24]. However, the inconvenience of colostomy and high morbidity rate, as well as functional limitations after APR, reduced

the patients' quality of life [25, 26]. In recent years, WLE has been gradually adopted as an alternative option because of its better functional performance, less invasiveness, and freedom from stoma than APR surgery [17, 26]. In addition, APR had a lower recurrence rate in local areas than WLE surgery, but no difference was noted in the 5-year survival rate in the recent study [9, 10, 17]. Therefore, the choice for surgery needs to consider the morbidity of the surgery and the patient's quality of life. Therefore, WLE may be another choice considering the patient's age, comorbidities, symptom control, and quality of life. The recent meta-analysis by Smith HG et al. reported that WLE with regular surveillance for local recurrence may be recommended as the primary strategy in most patients [27]. More studies revealed that WLE combined with adjuvant RT could not only reduce local recurrence than WLE alone but avoid the functional compromise compared with APR to solve the disadvantage of high recurrence in patients after WLE [9, 14]. Furthermore, sphincter-sparing WLE with RT had a similar local recurrent rate as APR [11, 15]. Moreover, Nusrath et al. [28] revealed that the better survival for rectal melanoma had been corrected using many factors, including the tumor size of < 2 cm and margin-negative resection without lymphovascular or perineural invasion. Debulking operation was performed first to decrease tumor volume to relieve symptoms and achieve consequent treatment with RT and WLE. In this case, RT and WLE not only had a striking response to residual tumor shrinkage but achieved margin-negative resection without severe side effects. In summary, no consensus exists on rectal melanoma treatment. In addition to traditional surgery and RT, immunotherapy is recently developing as an option [29, 30]. In conclusion, the current study presented a case with stage II rectal melanoma with a complete response by combining debulking surgery, RT, and WLE. For rectal melanoma that cannot be completely resected at the beginning and accompanied by bleeding symptoms, combining debulking surgery, RT, and WLE may be viable alternatives to provide the success rate of negative resection margins, lowered local recurrence rate, less medical cost, and better quality of life. However, a large-scale case study should be considered to validate its variability.

Abbreviations

RT: radiotherapy; WLE: wide local excision; MRI: magnetic resonance imaging; PET-CT: positron emission tomography-computed tomography; APR: abdominoperineal resection

Declarations

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Authors' contributions

All authors were involved in the preparation of this manuscript. Hao-Tse Chiu and Chia-Cheng Wen participated in the study design. Tung Liu participated in data collection. Hao-Tse Chiu participated in

literature searching. Hao-Tse Chiu and Chia-Cheng Wen wrote the manuscript. The authors read and approved the final manuscript.

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Availability of data and materials

The datasets obtained during the current study are available from the corresponding author on reasonable request.

Ethics approval and consent to participate

Not applicable.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and accompanying images

Competing interests

The authors declare no competing interests.

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Figures

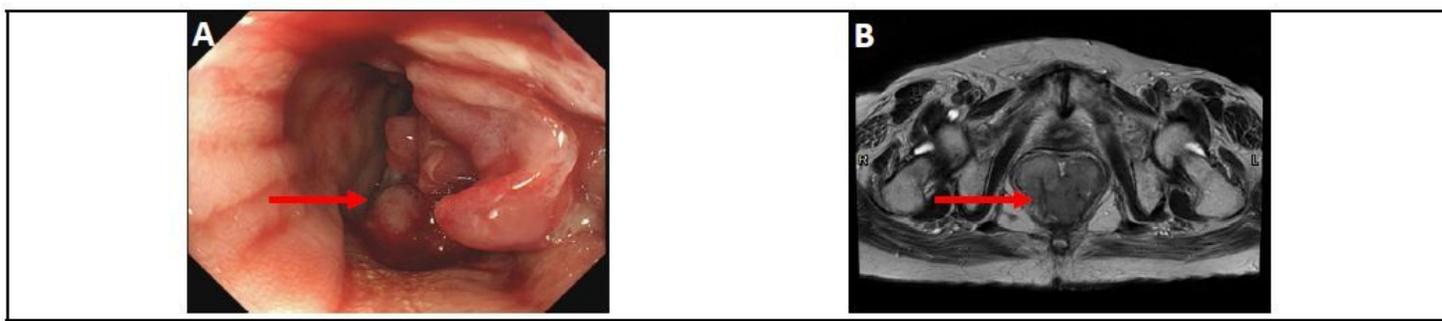


Figure 1

Initial colonoscopy and magnetic resonance imaging (MRI). A Colonoscopy showed a polypoid mass about 6 cm in size at the rectum. B MRI showed a lobulated mixed-signal intensity mass at the rectum (5.5 × 4.8 cm in size, 3.5 cm above the anal verge) on the T2-weighted axial image.

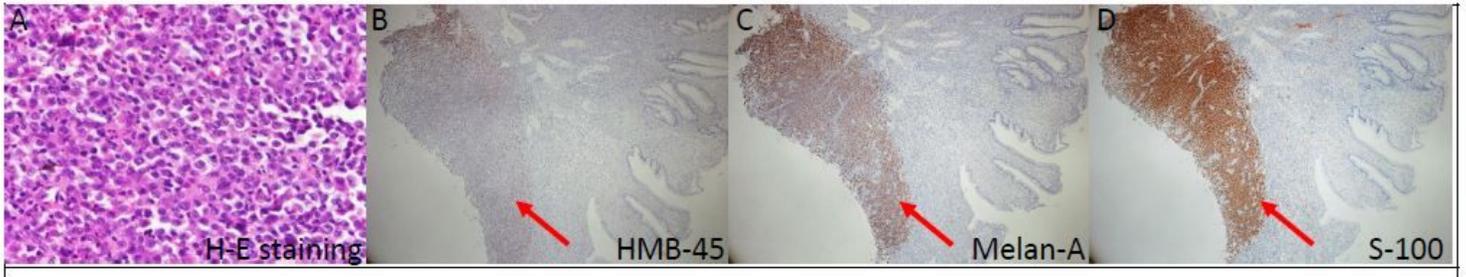


Figure 2

Histology image of the biopsied tissues A Neoplastic cells showed epithelioid morphology (hematoxylin-eosin staining, $\times 400$). B Neoplastic cells showed focally positive staining for HMB-45 (immunohistochemistry staining, $\times 40$). C Neoplastic cells showed positive staining for Melan-A (immunohistochemistry staining, $\times 40$). D Neoplastic cells showed positive staining for S-100 (immunohistochemistry staining, $\times 40$).

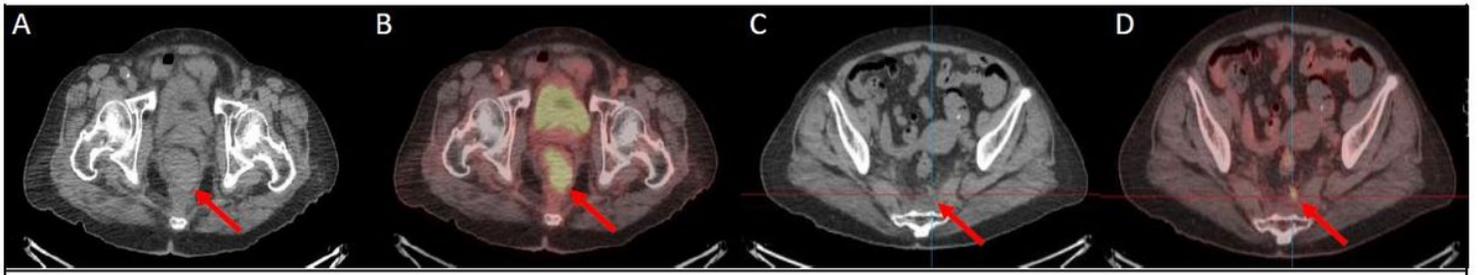


Figure 3

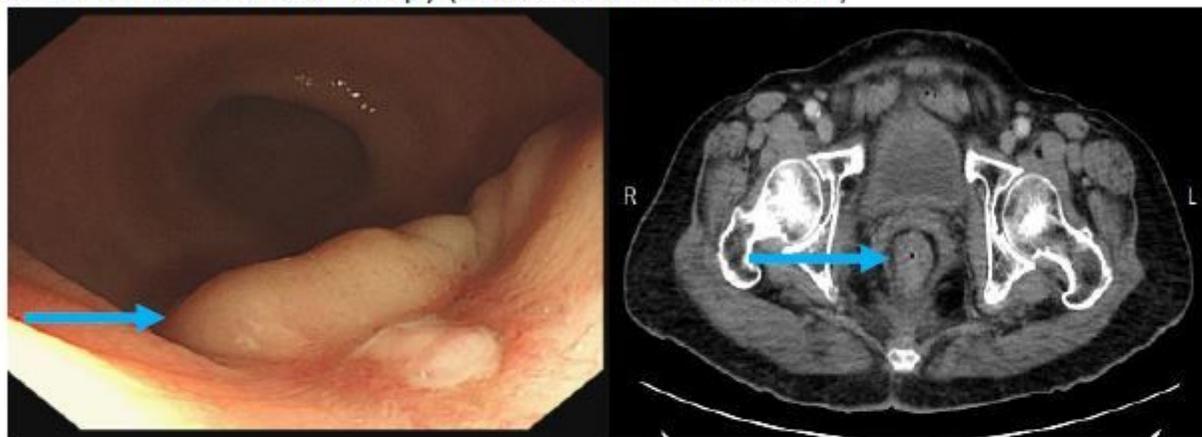
Computed tomography (CT) and positron emission tomography (PET/CT) for pelvic imaging in anorectal melanoma. The (A) axial CT and (B) PET/CT showed intense FDG-avidity over the rectum, suspicious for malignancy. The (C) axial CT and (D) PET/CT showed one mild FDG-avidity over the presacral space, suspicious for nodal metastasis.



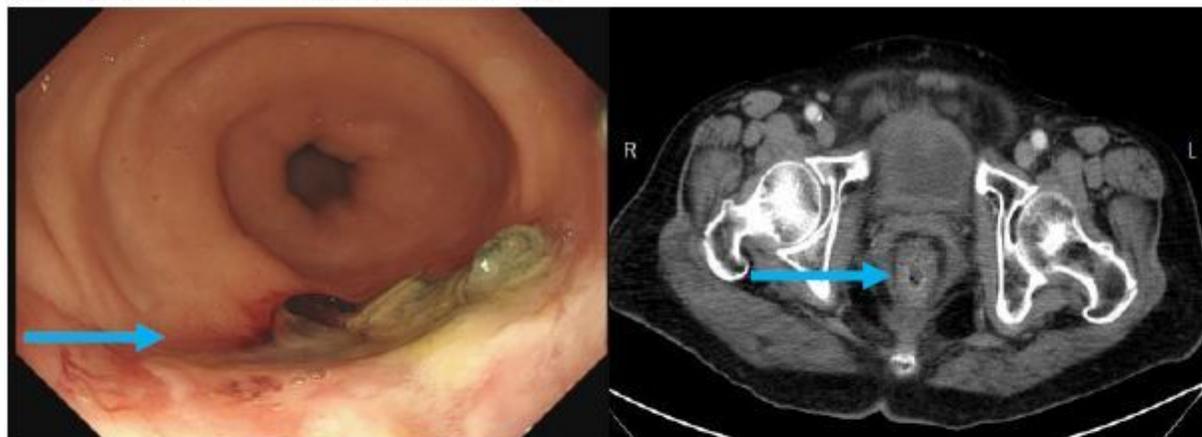
Figure 4

The piecemeal resection is done with snares to cut off parts of the polypoid mass piece by piece until the protruding mass is resected.

A 1 month after radiotherapy (before wide local excision)



B 3.5 months after wide local excision



C 9 months after wide local excision



Figure 5

Following-up colonoscopy and computed tomography (CT)