Multiple Synchronous Anorectal Malignant Melanoma Coexisting with Adenocarcinoma of the Sigmoid Colon

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Abstract Primary anorectal malignant melanoma is an exceptionally rare neoplasm associated with poor prognosis. Anorectal malignant melanoma has been very rarely described with coexisting primary tumors of the colorectum. A 56-year-old female patient was admitted with a history of rectal bleeding. She had experienced increasing constipation and a sense of obstruction in the rectum for 6 months. Flexible rectosigmoidoscopy showed a large, pedunculated polypoid lesion extending from the anal canal to the rectum. She underwent a transanal local excision and was diagnosed with a melanoma of the anorectum with positive margins. Therefore, a formal abdominoperineal resection was performed. In addition to multiple synchronous anorectal malignant melanoma, we incidentally found another primary tumor in the proximal surgical margin of the resected specimen. Histopathologically, the lesion was an intramucosal adenocarcinoma of the sigmoid colon. Postoperatively, the patient received adjuvant chemotherapy of six cycles duration. At present, the patient has completed 18 months of follow-up.

Keywords Malignant melanoma · Anorectal melanoma · Synchronous tumors · Abdominoperineal resection · Transanal local excision

Introduction

Anorectal region is the third most common site of melanoma after the skin and retina. Anorectal melanoma accounts for 0.2–3 % of all malignant melanoma cases and 0.1–4.6 % of malignant tumors of the rectum and anus [1]. The incidence of multiple primary cancers identified in the colon and rectum is estimated to be about 2 % to 11 % [2]. However, anorectal malignant melanoma has been very rarely described with coexisting primary tumors in the surgical specimens. We present a case of primary synchronous anorectal malignant melanoma concomitant with intramucosal adenocarcinoma of the sigmoid colon.

Case Report

A 56-year-old female patient was admitted with a history of recurrent rectal bleeding. The patient had experienced increasing constipation and a sense of obstruction in the rectum for 6 months. The patient had also noticed a lump at the anus. There was not loss of weight. The patient had undergone a gynaecological operation and an appendectomy 20 years earlier in her past medical history. The family and social histories were noncontributory.

On admission, physical examination was normal. Inguinal examination revealed no regional lymphadenopathy. Digital examination of the rectum revealed multiple, asymmetrical, firm, and nodular lesions, the largest being 3 cm in diameter, in the posterior wall of the rectum filling the

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lumen, the lower limit extending down to the level of the dentate line. No abnormalities were found by routine laboratory examinations. The levels of carcinoembryonic antigen (CEA) was 5.52 ng/mL (normal range: 0–4.3 ng/mL).

Abdominopelvic computed tomography (CT) with oral and intravenous contrast demonstrated an annular lesion with irregular wall thickening along a 5-cm segment in the distal rectum and perirectal enlarged lymph nodes. No luminal growth was detected in the descending and sigmoid colon at the CT slices. Positron emission tomography (PET) scan did not detect the evidence of metastatic disease.

Flexible rectosigmoidoscopy showed a large, pedunculated polypoid lesion extending from the anal canal to the rectum, but a biopsy was not taken because of it could be a vascular lesion in origin or thrombosed hemorrhoid. The mucosa was normal, and no luminal lesion was detected in the descending and sigmoid colon. Hereupon, we performed a rectal examination under general anesthesia, and the anorectal tumorous mass was excised locally. Pathologic examination of the lesion revealed malignant melanoma with positive surgical margins. Immunohistochemical stainings of the tumor cells were positive for HMB-45, Melan A and S-100 protein. Therefore, a formal abdominoperineal resection (APR) of the rectum was performed.

Macroscopically, on a 29-cm-long terminal colon specimen along with perianal skin, three polypoid tumoral masses were found in the anorectum: (1) $3.0 \times 2.0 \times 1.0$ cm, in the dentate line, (2) $3.3 \times 2.5 \times 2.0$ cm, 1 cm proximal to the dentate line, (3) $2.0 \times 1.5 \times 0.5$ cm, 4 cm proximal to the dentate line (Fig. 1 a–b). Between described tumors there were areas of normal rectal mucosa without tumor involvement. The largest tumor (no.2) penetrated through the entire rectal wall and reached the perirectal fat. Metastatic tumor was found in 16 of 27 regional lymph nodes. In addition to multiple synchronous anorectal malignant melanoma, we incidentally found another primary tumor, which was 0.6×0.5 cm in size and located 5 cm away from the proximal surgical margin of the resected specimen. Histopathologically, the lesion was an intramucosal adenocarcinoma (Fig. 2 a). Positive p53 nuclear staining was observed (Fig. 2 b). The tumor had stromal invasion. The patient was discharged from our department 2 weeks after the operation following a non-eventful course.

Since metastatic pulmonary nodules were detected on the follow-up thoracic CT scans, the first-line chemotherapy for metastatic melanoma was started with dacarbazine (DTIC; dimethyl triazenochlorethyl nitrosourea) 200 mg/m^2 intravenously for 2 days of the week and cisplatin 35 mg/m^2 intravenously for 3 days of the week. The patient received 6 cycles of chemotherapy. She has been well for more than 18 months since her operation.

Fig. 1 a–b Gross findings of the resected specimen. Multiple blackish tumors were observed in the anorectal region: one in the dentate line (no.1), the other one at 1 cm proximal to the dentate line (no.2), and the last one at 4 cm proximal to the dentate line (no.3)

Discussion

We wrote this paper as a case report because of the relative rarity of primary malignant melanoma of the anorectal region. There have been over 500 cases of this uncommon anorectal malignancy reported in the literature to date.

Fig. 2 Intramucosal adenocarcinoma of the sigmoid colon (H&E, $\times 200$) (a). Positive immunohistochemical staining for p53 ($\times 200$) (b)

Anorectal malignant melanoma has been described with coexisting primary adenocarcinoma or sarcoma in the surgical specimens. A search of the literature uncovered 4 cases of coexistent malignant melanoma and adenocarcinoma or sarcoma in the anorectum and colon [3–6]. Secco et al. reported a similar case of malignant melanoma in the anorectal junction synchronous with adenocarcinoma of the sigmoid colon [6].

Malignant melanoma of the rectum is relatively rare, accounting for 0.8 %–1 % of all colorectal neoplasms and 0.7 % of all malignant melanomas at any site [7]. Rectal melanomas are more aggressive and their biological behavior is different from that of melanoma of the skin. Long-term survival is very low; because of early systemic spread, only 6.7 % to 12 % of patients are reported to be free of the disease 5 years after the operation [8]. Differential diagnosis includes carcinoma of the rectum, prolapsing rectal polyp, and thrombosed external piles. To establish a definitive diagnosis of anorectal melanoma from a preoperative biopsy specimen is quite difficult and therefore, it is occasionally misdiagnosed as a different type of neoplasm. We made a pathological diagnosis of malignant melanoma with a positive surgical margin after performing a rectal examination and local excision under general anesthesia.

There is no consensus at this moment on which surgical approach is preferred. However, there are two opinions concerning the surgical treatment of anorectal malignant melanoma. First, wide local excision should be considered a procedure of choice, because of the high morbidity and mortality rate, and lack of survival advantage of associated with APR. Second, APR can improve locoregional control rates, which can improve the good prognosis. Because transanal local excision has less morbidity than APR and avoids the necessity of a colostomy, most authors consider it as the first choice of treatment for anorectal malignant

melanoma when it is technically feasible. Therefore, we initially performed wide local excision. Since the surgical margins were positive, then APR was performed in our patient.

In summary, we have reported a case of multiple synchronous primary melanoma of the anorectal region with a coexistent primary adenocarcinoma of the sigmoid colon. Because the disease stage appears to be more important than the type of operation, it is necessary, in parallel with attempts for treatment methods, to promote an awareness of the importance of an early diagnosis.

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