Case Report

Head Subcutaneous Metastasis of Endocrine Cell Carcinoma in the Rectum

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Abstract

A 46-year-old man who presented with rectal bleeding was diagnosed with rectal carcinoma. As the tumor was found within 2 cm of the anal verge, he underwent abdomino-perineal resection in November, 2001. The invasive front of the tumor was within the wall of the rectum without serosal invasion. The surgical specimen was diagnosed as endocrine cell carcinoma and one regional lymph node metastasis was revealed located near the tumor. After discharge, the patient underwent adjuvant chemotherapy consisting of p.o. 5-fluorouracil 200 mg/day for more than 6 months. He noticed a head subcutaneous tumor in May, 2002. In June, two more tumors appeared. The tumors were removed surgically under local anesthesia. The surgical specimens were diagnosed as endocrine cell carcinoma, and were judged to be metastasis from the rectal lesion, based on the histological similarity. Cutaneous metastasis of rectal adenocarcinoma is a rare event occurring in fewer than 4% of all patients with rectal cancer. Cutaneous metastasis of endocrine cell carcinoma in the rectum is extremely rare. Furthermore, to our knowledge, no head subcutaneous metastasis of endocrine cell carcinoma in the rectum has been reported to date.

Key words Endocrine cell carcinoma, Head subcutaneous metastasis irinotecan

Introduction

In the recently released World Health Organization classification of gastroenteropancreatic (GEP) endocrine tumor,1 poorly differentiated endocrine carcinomas (PDECs) constitutes a distinct category separate from the other two main categories (i.e. well-differentiated endocrine tumors and well-differentiated endocrine carcinomas) because of the distinctive histological features and highly aggressive clinical behavior with poor prognosis.2 However it is extremely unusual to detect head subcutaneous metastasis from a rectal lesion even if the case is in the terminal stage. In this report we describe a case of PDEC of the rectum of which the first recurrence lesion was his head subcutaneous, with a relatively long interval between the appearance of subcutaneous metastasis and death.

Case Report

A 46-year-old man who presented with rectal bleeding was diagnosed with rectal carcinoma. As his tumor was found within 2 cm of the anal verge, he underwent abdomino-perineal resection in November, 2001 (Fig. 1). The invasive front of the tumor was within the wall of the rectum without serosal invasion. The surgical specimen was diagnosed as endocrine cell carcinoma by pathological examination including grimeilus and neuron specific enolase (NSE) immunolabeling...
of the histological features (Fig. 2A, B, C) and revealed one regional lymph node metastasis located near the tumor. Ki 67 immunolabeling of the histological features showed that the tumor was low grade malignancy (Fig. 2D).

After discharge, he underwent adjuvant chemotherapy consisting of p.o. 5-fluorouracil 200 mg/day for more than 6 months. He noticed a head subcutaneous tumor in May, 2002. In June, two more appeared (Fig. 3). The tumors were removed surgically under local anesthesia. The surgical specimens were diagnosed as endocrine cell carcinoma by pathological examination including grimelius and neuron specific enolase (NSE) immunolabeling of the histological features (Fig. 4A, B, C), and were judged to be metastasis from rectal lesions, based on the histological similarity. Ki 67 immunolabeling of histological features showed that the metastatic lesions were high-grade malignancy (Fig. 4D). We performed computed tomography (CT) and radioisotope examination, but no other was detected. In addition, the blood serotonin level was within normal limits.

Therefore, 5-fluorouracil was not an effective agent for his disease. He underwent systemic chemotherapy consisting of irinotecan (CPT-11) 80 mg/m² every 2 weeks from August, 2002. However, we confirmed multiple spreading to the lungs by CT in February, 2004. In addition,
cervical spine metastasis was diagnosed by radioisotope examination. He died in September, 2004, which is more than 24 months after systemic chemotherapy was initiated, and approximately 36 months after the first operation was performed.

Discussion

Cutaneous metastasis of rectal adenocarcinoma is a rare event occurring in fewer than 4% of all patients with rectal cancer. While metastasis can be found in any location of the cutaneous, rectal cancer most often metastasizes to the middle or lower dermis of the abdomen and the perianal skin, usually to the abdominal wall and around the umbilicus; the metastasis also often appear after surgery for rectal carcinoma.

On the other hand, endocrine cell carcinoma most often metastasizes to liver and systemic lymph nodes. Cutaneous metastasis of endocrine cell carcinoma in the rectum is extremely rare. Furthermore, to our knowledge, no cases of head subcutaneous metastasis of endocrine cell carcinoma in the rectum have been reported.

As to Ki 67 immunolabeling of histological features, the primary lesion was low-grade malignancy, but the metastatic lesions were high-grade malignancy. In our patient, there was no relationship between the grade of malignancy in
the primary lesion and the occurrence of distant metastasis.

With regard to adjuvant chemotherapy for endocrine cell carcinoma, some reports state that 5-fluorouracil is an effective agent for liver metastasis.6 However, our adjuvant chemotherapy consisted of 5-fluorouracil 200 mg/day per os, and head subcutaneous metastasis occurred within 1 year after the first operation. No other effective chemotherapeutic drugs for endocrine cell carcinoma have been reported. However, there was one report that irinotecan is an effective agent for pulmonary small-cell carcinoma.7 Therefore, we administered irinotecan after removing the head lesions.

Survival after diagnosis of skin metastasis ranges from 1 to 34 months.8,9 Our patient survived for more than 24 months from diagnosis of subcutaneous metastasis to death, which is a relatively long survival following diagnosis of subcutaneous metastasis. Although irinotecan might be useful as chemotherapy for endocrine cell carcinoma, we have no evidence to support this.

As to the metastatic route to head subcutaneous, it may be either by hematogenous or lymphovascular spread.

References