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Citation	日本外科宝函 (1983), 52(5): 710-717
Issue Date	1983-09-01
URL	http://hdl.handle.net/2433/208880
Right	
Type	Departmental Bulletin Paper
Textversion	publisher

Arch Jpn Chir 52(5), 710～717, Sept., 1983

A Case of Hodgkin's Disease of the Rectum

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Received for Publication June. 17, 1983.

Abstract

Hodgkin's disease solitarily involving the rectum is quite an uncommon condition which has rarely been reported heretofore. Recently, a patient with this disease was encountered.

A 68-year-old Japanese men with the chief complaints of anal pain and constipation visited our clinic on November 16, 1981. Rectal examination disclosed a palpable mass of Borrman II type in the anterior rectal wall. After radiotherapy with ^{60}Co, a Miles operation was perfomed. Radiotherapy was also carried out, combined with BONP therapy in the postoperative stage. The patient was discharged on March 14, 1982. He was successfully rehabilitated. Histopathologically, many Reed-Sternberg cells were found in the ulcer floor.

To the best of our knowledge, the present case was the first report in Japan. An effective treatment of this disease that promises favorable results seems to be surgical resection combined with radiotherapy and combined antitumor drug therapy.

Introduction

Hodgkin's disease of rectal origin is a very rare condition that has so far been reported only infrequently. Recently, we had experience with a patient with Hodgkin's disease involving the rectum alone, who had been erroneously diagnosed as having rectal cancer for lack of symptoms such as general lymph-node swelling, hepatomegaly, splenomegaly, and eosinophilia, which are usually characteristic of the disease. The present paper discribes the findings in this particular case and some information obtained by literature review.

Report of a Case

A 68-year-old man with anal pain and constipation was first see in November 1981. He had suffered from internal hemorrhoids for many years previously, however, in October 1980, anal pain was enhanced, and he became constipative and had a weight loss of about 11 pounds.

Physical findings on examination: His physical constitution was moderate. He was well

Key Word: Hodgkin's disease of the rectum, Reed-Sternberg cells.
索引語：直腸，ホジシン病，リードステルンベルグ細胞.
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The gist of the present paper was presented at the 37th annual meeting of the Japan Socitely of Colo-Proctology (Tokyo, November, 1982).

Table 1. Laboratory findings on admission

(1)	Hematologic findings RBC: 501×10^4/mm^3, Hb: 18.0 g/dl, Ht; 54%, WBC: 7,000/mm^3 (eosinophils: 1%, basophils: 3%, lymphocytes: 29%), and platelet: 20.0×10^4/mm^3
(2)	Blood biochemical findngs Total protein: 7.0 g/dl, albumin: 3.8 g/dl, GOT: 22, GPT: 17, LDH: 146, alkaline phosphatase: 62, and CEA: 1.7 ng/ml

nourished and was neither anemic nor jaundiced. None of the cervical, maxillary, and inguinal lymph nodes were enlarged. The heart and lung were normal. He had a flat belly, and the liver, spleen, or kindneys were not palpable. Rectal examination was performed and, as a result, a concave mass involving primarily the anterior wall of the rectum was palpated and the lumen in that part of the organ was narrowed.

Examination data on admission (Table 1): Hematologically, anemia was unseen. Atypical lymphocytes were not detected, nor was lymphopenia or eosinophilia evidenced. Liver function tests were all within normal limits. The only abnormality was positive occult blood.

Simple chest *x*-ray examination showed that lymph nodes in either the mediastinal or hilar region were not swollen, and pulmonary markings were normal (Fig. 1)

Ultrasonographic examination of the abdomen revealed that ascites and liver metastasis were absent, and that the spleen and kidneys were normal.

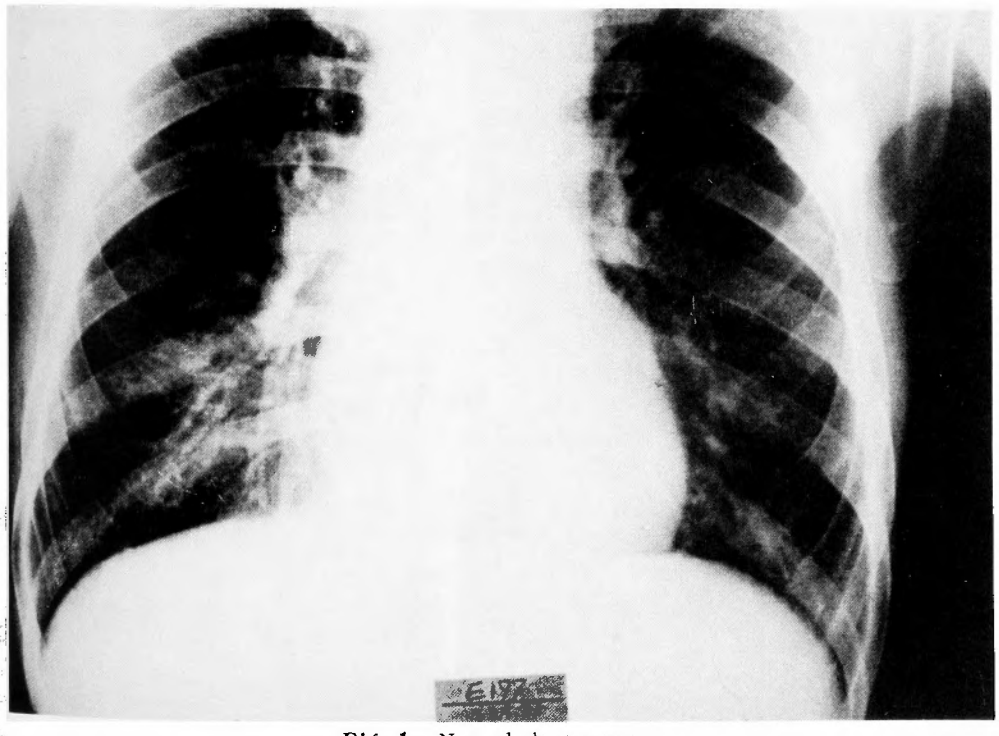

Fig. 1. Normal chest x-ray

Barium-enema examination showed not only an irregularly bordered filling defect approximately 3 cm from the anal verge but also a narrowing of the rectal lumen (Fig. 2).

Rectoscopically, a Borrmann II type tumor covering one-third of the circumference of the rectum was recognized. Its center was about 5 cm from the anal verge.

Preoperative biopsy demonstrated not only multinucleated cells and lymphocytes, but also large atypical mononucleated cells, some of which were binucleated, in the granulation tissue infiltrated with plasmocytes (Fig. 3). In some areas, high-density shadows indicating intensive eosinophilic infiltrations were recognized. The intranuclear structures were unclear because of strong artifacts and a diagnosis of undifferentiated carcinoma was made.

Preoperative treatment: Based on the principle of rectal cancer treatment in our department, radiotherapy was performed preoperatively. At the time of initial examination, the tumor protruded into the lumen of the rectum as ascertained by rectal examination. As therapy progressed, the protrusion regressed and the lesion looked as if it were ulceration. When the radiation dose amounted to 1,400 rads, the symptoms of fever and pain were exacerbated. Therefore, therapy was discontinued and a Miles abdominoperineal resection was performed.

Operative findings: No ascites was present in the peritoneal cavity, nor was there a metastatic lesion in the liver. Swollen lymph nodes (Azuki-bean-sized) were found at the root of the inferior mesenteric artery, but lymph nodes in all other regions, including preaortic ones were normal. The tumor mass existed in the anterior wall of the rectum, but it partially penetrated into the prostate. Miles abdominoperineal resection was performed and, simultaneously, evide-

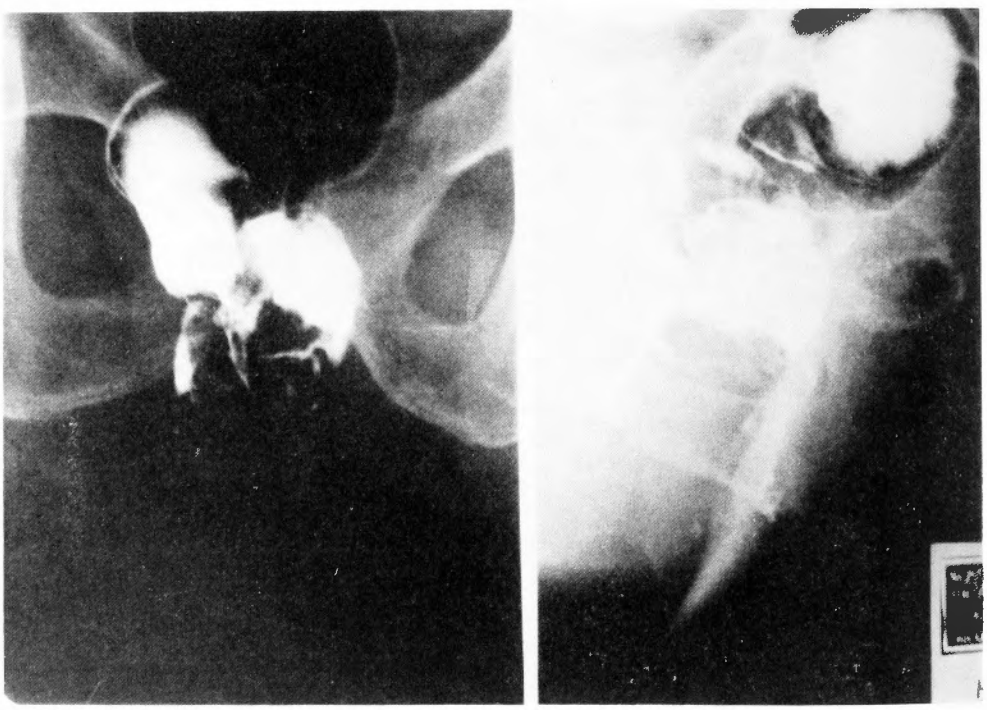

Fig. 2. Barium enema (left: front view, right: side view).

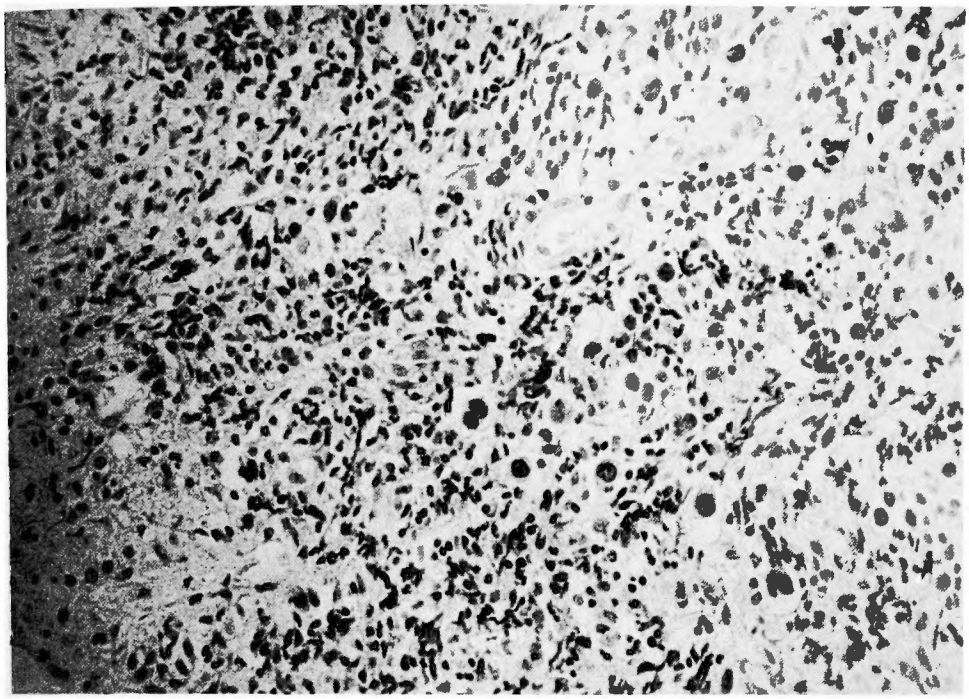

Fig. 3. Histology of preoperative biopsy (hematoxylin and eosin; ×125).

Fig. 4. Cut surface of resected specimen.

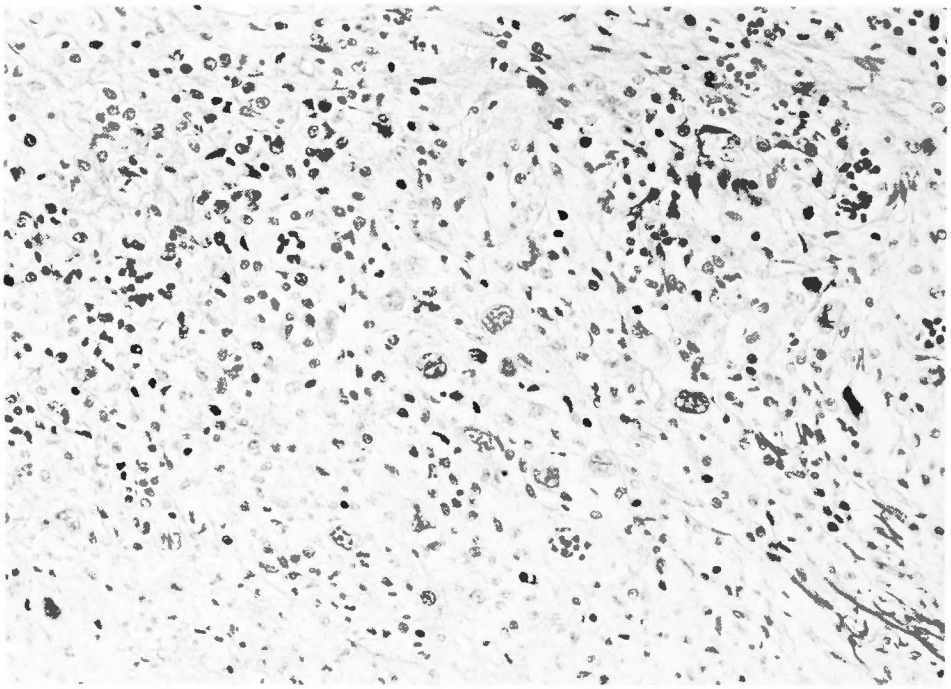

Fig. 5. Microscopic findings of resected apecimen (hematoxylin and eosin; ×250).

ment of the related lymph nodes was done.

Macroscopic findings of the resected specimen: Serial rectangular sections of the rectal wall were prepared. Figure 4 shows one of those sections from the anal side. A round ulcer, 2.5×2.5×0.5 cm in size, was present 3 cm from the dentate line on its oral side. The base of the lesion was flat, up to the periphery. The lesion was a type IV benign ulcer which was not protuberant and instead penetrated into the prostate.

Histopathologic findings: Large atypical mononucleated cells possessing nuclei, which are similar to the bubble-shaped nuclei with clear nucleoli of binucleated or multinucleated Reed-Sternberg cells, appeared in the granulation tissue of the ulcer base. In view of the fact that

Table 2. Particulars of treatments

Preoperative treatment:
Radiotherapy with 1,400 rad (7×200 rad) ^{60}Co
Surgical treatment:
Miles abdomnoperineal resection (R_3)
Postoperative treatment:
Radiotherapy with 3,000 rad (15×200 rad) ^{60}Co
+
BONP therapy with 15 mg/week of bleomycin, 1 mg/week of vincristine, 50 mg/week of procarbazine, and 30 mg/day of prednisolone
+
OK-432 administration (maintenance dose: 2KE)

coexistent lymphocytes were not atypical, the patient was diagnosed as having Hodgkin's disease. The lymph nodes removed were proved intact and tumor cells were localized within the ulcer floor (Fig. 5).

Postoperative treatment: Radiotherapy (3, 000 rads) and multiple antitumor chemotherapy, which was composed of BONP therapy (15 mg/week of bleomycin, 1 mg/week of vincristine, 50 mg/day of procarbazine, and 30 mg/day of prednisolone) and OK-432 treatment were carried out after operation (Table 2).

The patient was discharged on March 14, 1982. Since then, he has been well with no signs of recurrence.

Discussion

Hodgkin's disease often involves the digestive tract. In most cases, however, this occurs in an advanced stage of the disease after systemic dissemination[2] and the predilection for it is the terminal portion of the ileum where Peyer's patches are well developed[6] In 1907, HESS et *al.*[8] reported multiple Hodgkin's granuloma involving the stomach, small intestine, and rectosigmoid flexure. In 1956, GECHMAN *et al.*[6] reviewed the literature and found four cases of Hodgkin's disease of the rectum. Since then, one case of Hodgkin's disease of the rectum has been reported by SHAPIRO *et al.* (1961)[14], two by PERRY *et al.* (1972)[13], and another by HARNED *et al.* (1976)[7]. These eight cases include only one patient in whom the lesion was limited to the rectum and none of the systmic lymph nodes were swollen. When the nine cases including the present case were classified by sex, the ratio of men to women was 1 to 1. 3. Thus, women were slightly more susceptible to the disease. Their ages ranged from 22 to 68 years with an average of 40. 3 years (Table 3).

The pathogenesis of Hodgkin's disease of the digestive tract remains to be clarified. However, due to the fact that these organs are involved mainly in a late stage of the disease, an inference may be drawn, as follows: The disease first affects the mesenteric lymph nodes, then lymphotropic metastasis occurs retrogressively to involve the digestive tract[9]. However, in many of the reported cases of Hodgkin's disease of the rectum, the organ was invaded via the retroperitoneal route. The present case was unique in that the tumor developed solitarily in the rectum and any abnormalities were unseen in the related areas., and the pathogenesis was

Table 3. Reported cases of Hodgkin's disease of the rectum (1938-1983)

No.	Authors	Age (yrs)	Sex	Lymph nodes & other organs involved
1	Gallart, et al. (1938)	35	F	inguinal, spleen, liver
2	Craver, et al. (1945)	22	F	cervical, axillary
3	Pettinari, et al. (1947)	40	M	no signs of dissemination
4	Gechman, et al. (1956)	40	F	cervical, spleen, liver
5	Shapiro, et al. (1961)	46	M	cervical, axillary, inguinal, liver, spleen
6	Harned, et al. (1976)	31	M	axillary, inguinal, liver, spleen
7	Present authors	68	M	no signs of dissemination

unclear in this case.

Symptoms associated with Hodgkin's disease of the rectum, in common with other malignancies involving this organ, are due primarily to obstruction[15]; that is, local pain, change in bowel habit, bleeding, and palpability of a mass. They may be accompanied with general symptoms such as Pel-Ebstein fever, pruritus, loss of weight, and anemia; however, anemia is rarely seen in Hodgkin's disease of the digestive tract[6].

Diagnostic means comprise barium-enema examination, rectoscopy, rectal fiberoscopy, and biopsy. It is true that the rectal wall in a focal region is somewhat softer in Hodgkin's disease than in cancer, but it is almost impossible to differentiate the former from the latter solely by barium enema examination[3,7] In order to establish a diagnosis of Hodgkin's disease by biopsy, it is a requisite to confirm the following two points: 1) peculiar Reed-Sternberg cells are present, 2) coexistent lymphocytes are not atypical[5,11]

Chemotherapy for Hodgkin's disease is performed mainly by means of MOPP* treatment[9]. In the MOPP treatment, the CR** rate is usually as high as 80 per cent and may increase to 100 per cent in those patients without general symptoms[9]. In the present study, however, we selected BONP treatment, in which bleomycin is substituted for NH_2.

The diagnostic criteria of lymphoma of large intestinal origin, which were proposed by DAWSON *et al.*[4], are briefly described, as follows: 1) superficial lymph nodes are not palpable at the time of the first examination, 2) chest *x*-ray examination shows that the mediastinal lymph nodes are not swollen, 3) WBC and leukocyte differential count are normal, 4) at the time of laparotomy, only lymph nodes around a tumor are found to be swollen, and 5) neither the liver nor the spleen is invaded by tumor cells. According to the above criteria, our patient was considered to have Hodgkin's disease of rectal origin.

Furthermore, this case comes under the category of stage I_E according to Hodgkin's disease staging classification of Ann Arbor[1] or classified under the category of H_0, Po, No, Ai, Iyo, Vo (stage III) according to the rules for interpretation of large intestinal cancer in Japan[10]. It seems that the clinical stage of Hodgkin's disease of rectal origin is better represented by the latter classification. This disease is considered to be of "mixed cellularity" (MC) type according to the classification of Rye (1966)[12].

Conclusion

We report a case of Hodgkin's disease of rectal origin to demonstrate that Hodgkin's disease as well as carcinoma and non-Hodgkin's lymphoma may possibly develop in the rectum. The characteristics of Hodgkin's disease of the rectum are discussed, referring to the literature.

* a regimen of mechlorethamine, Oncovin (vincristine), procarbazine, and prednisone, used in cancer chemotherapy.
** Complete Regression

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和文抄録

直腸ホジキン病の一例

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玄　　栄　世

直腸原発のホジキン病は極めて稀な疾患であり，文献的にも散見するにすぎない．我々は，最近，68才男性で，肛門部痛および便秘を主訴として来院し，直腸指診で前壁に，Borrman II型の腫瘤を触知し，術前^{60}Co放射線療法施行後，直腸切断情を施行し，術後にも，放射線療法と多剤併用化学療法（MOMP）を施行し，元気に社会復帰せしめ得た症例を経験した．本症例は，本邦第1例目と思われ，本症は，直腸に，非ホジキンリンパ腫と同様に存在するものであり，その治療法は，外科的切除か，放射線療法が有力であり，さらに多剤併用による免疫化学療法との合併治療が，良好な結果をもたらすと考えた．