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# Laparoscopic surgery for colon cancer with intestinal malrotation in adults: Two case reports and review of literatures in Japan

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## ABSTRACT

**INTRODUCTION:** Intestinal malrotation is a congenital anomaly, and its occurrence in adults is rare. Colon cancer with intestinal malrotation is far more rare. We herein report two cases of colon cancer with intestinal malrotation treated with laparoscopic surgery and reviewed the literatures in Japan.

**PRESENTATION OF CASES:** Case 1 involved a 78-year-old man. Abdominal enhanced computed tomography (CT) showed that the tumor was located in the sigmoid colon. Intraoperatively, the cecum and ascending colon were located along the midline and the small intestine occupied the right side of the abdomen. The tumor was located in the cecum, and the patient was diagnosed with cecal cancer with intestinal malrotation. We performed laparoscopy-assisted ileocecal resection. Case 2 involved a 81-year-old man. Colonoscopy revealed a laterally spreading tumor in the cecum. Intraoperatively, the position of the small intestine and the ascending colon was similar to case 1, and Ladd's band was found in front of the duodenum. Thus, we diagnosed the patient with a laterally spreading cecal tumor with intestinal malrotation and performed laparoscopy-assisted ileocecal resection.

**DISCUSSION:** A review of the literature revealed 49 cases of colon cancer with intestinal malrotation and laparoscopic surgery performed at 30.6%. If laparoscopic mesenteric excision for colon cancer with intestinal malrotation is unsafe because of the abnormalities of the artery, mesenteric excision should be performed outside the body.

**CONCLUSION:** If the intestinal malrotation is diagnosed preoperatively, 3D-CT angiography should be used to reveal the vascular anatomic anomalies for safe performance of laparoscopic surgery.

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## 1. Introduction

Intestinal malrotation is a congenital anomaly that may cause intestinal obstruction or midgut volvulus in infants. The diagnosis of intestinal malrotation in adults is rare because most patients remain asymptomatic.

The incidence of colorectal cancer has gradually increased. In 2016 in Japan, this cancer was ranked as the second and fourth most common type among women and men, respectively [1]. The laparoscopic approach for colon cancer has recently become a practical technique, but the optimal surgical procedure for treatment of colon cancer with intestinal malrotation has not been established because of the rarity of intestinal malrotation.

We herein report two cases of laparoscopic surgery for colon cancer with intestinal malrotation in adults and reviewed the literatures in Japan.

This case report is compliant with the SCARE Guidelines [2].

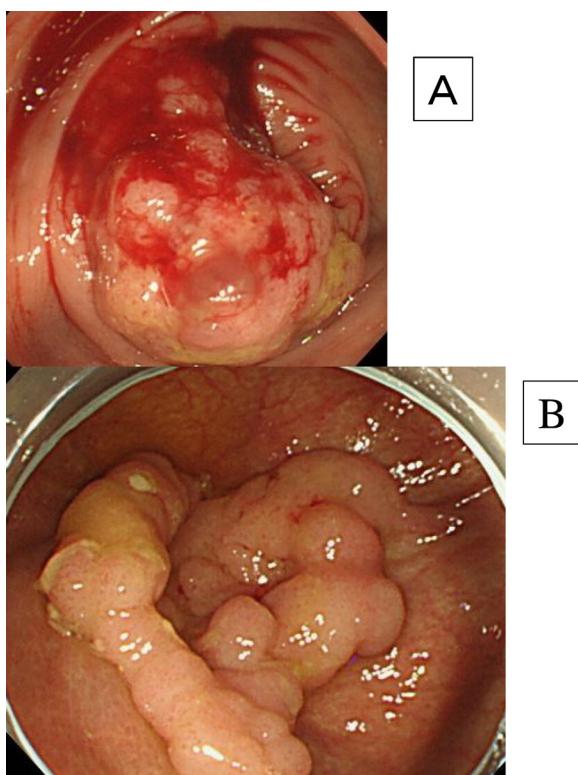
## 2. Presentation of cases

### 2.1. Case 1

A 78-year-old man visited our clinic because of constipation. Colonoscopy revealed a type II tumor located 50 cm from the anal verge (Fig. 1A). Abdominal enhanced computed tomography (CT) showed that the tumor had thick walls and was located in the center of the abdomen without lymph node swelling or metastatic lesions. From these findings, we diagnosed the patient with sigmoid colon cancer preoperatively. Intestinal malrotation was not suspected preoperatively, but a subsequent review of the imaging study demonstrated that the superior mesenteric vein (SMV) was located on the left side of the superior mesenteric artery (SMA) (Fig. 2A). We scheduled laparoscopy-assisted sigmoid colectomy. Intraoperative examination revealed that the small intestine was

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**Fig. 1.** (A) In Case 1, colonoscopy showed a type II tumor located 50 cm from the anal verge. (B) In Case 2, colonoscopy showed a laterally spreading tumor located in the cecum.

occupied the right side of the abdomen. The ileocecal region was located along the midline of the abdomen, and the marking for the tumor was found in the ascending colon. The ascending colon and cecum were not fixed with the retroperitoneum, and the ligament of Treitz could not be clearly identified. The patient was diagnosed with cecal cancer with intestinal malrotation (nonrotation type) (Fig. 3A, B). Laparoscopic mesenteric excision was considered unsafe because of the vascular and lymphatic anomalies. After mobilization of the ascending colon from the transverse colon, ileocecal region take out outside body from umbilical wound and mesenteric excision was performed outside the body. Because of the abnormalities of the artery, it was unsafe to perform right hemicolectomy with D3 lymph node dissection. Finally, we performed the ileocecal resection with D1 lymph node dissection. We considered D1 lymph node dissection was not adequate oncolog-

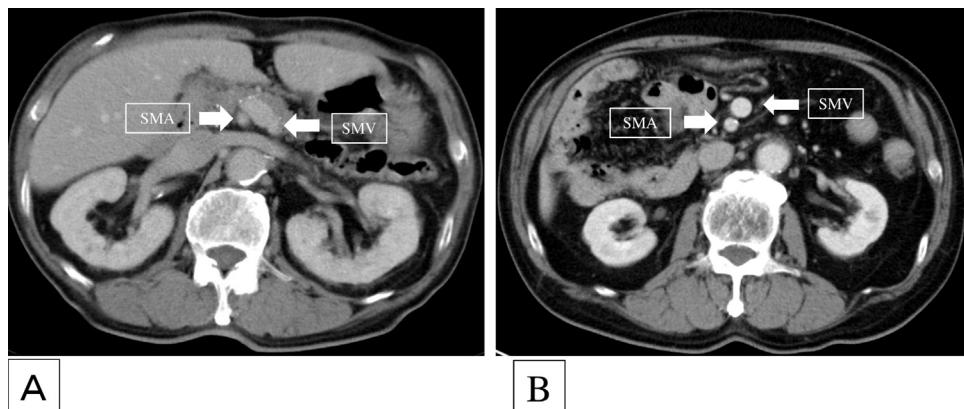
ically. Histopathological examination revealed well-differentiated tubular adenocarcinoma of the cecum infiltrating the subserosal layer without lymph node metastasis (pT3N0M0 = pStageIIA). Post-operative adjuvant chemotherapy was not performed and he has followed without recurrence for 5 years.

## 2.2. Case 2

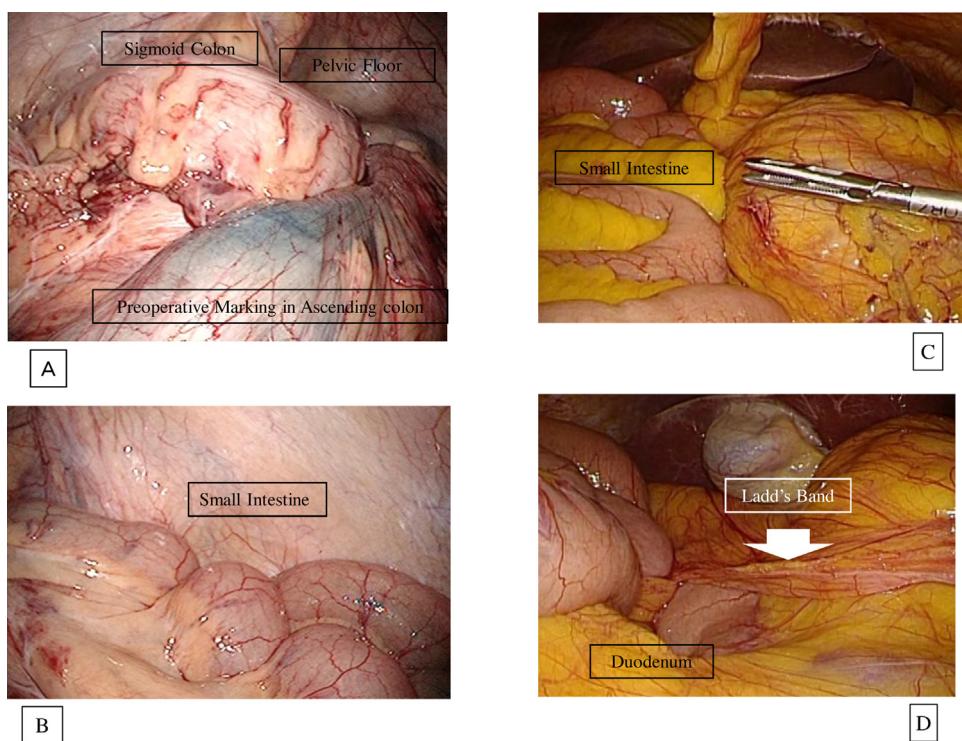
A 81-year-old man visited another hospital because of fecal occult blood. Colonoscopy revealed a laterally spreading tumor in the cecum (Fig. 1B). Abdominal enhanced CT showed that the tumor was located in the center of the abdomen. No lymph node swellings or metastases were present. Intestinal malrotation was not suspected preoperatively. However, a retrospective review of the CT image demonstrated that the SMV was located on the left side of the SMA and that the small intestine and colon occupied the right and left sides of the abdominal cavity (Fig. 2B). These signs were identical to those in Case 1. We scheduled laparoscopy-assisted ileocecal resection. Intraoperative examination revealed that the omentum was extensively adhered to the right wall of the abdomen. Upon peeling off this adhesion, the small intestine was found to occupy the right side of the abdomen. A further search of the intraperitoneal region showed that Ladd's bands were lying in front of the duodenojejunal junction, and the duodenum (which was free from the retroperitoneum) passed straight down to join the jejunum to right upper quadrant (Fig. 3C, D). We diagnosed the patient with a laterally spreading cecal tumor with intestinal malrotation (nonrotation type). The adhesion between the ascending colon and transverse colon was exfoliated by sharp dissection. After mobilization of the ascending colon, lymphadenectomy was performed outside the body because of the vascular and lymphatic anomalies. Finally, we performed ileocecal resection with D1 lymph node dissection. Histopathological examination revealed well-differentiated tubular adenocarcinoma of the cecum infiltrating the mucosal layer without lymph node metastasis.

## 3. Discussion

The midgut rotates 270° counterclockwise around the SMA and is fixed to the retroperitoneum at 4–12 weeks of fetal life. The process of rotation has been conveniently divided into three stages [3]. The first stage is essentially that of an umbilical loop with two limbs lying beside one another, the second is the stage of beginning of intestinal rotation, and the third is the stage of fixation of the intestine and fusion of its mesentery. Intestinal malrotation is defined faulty rotation with fixation of the midgut. In several reports, the various forms of intestinal malrotation has been clas-



**Fig. 2.** (A, B) Abdominal enhanced computed tomography showed that the superior mesenteric vein (SMV) was located on the left side of the superior mesenteric artery (SMA) (SMV rotation sign) in both cases.



**Fig. 3.** (A, C) Intraoperative examination revealed that the small intestine occupied the right side of the abdomen in both cases. (B) In Case 1, the ileocecal region was located along the midline of the abdomen, and the preoperative marking for the tumor was found in the ascending colon. (D) In Case 2, Ladd's bands were lying in front of the duodenum.

sified [4–6]. Wang and Welch [5] classified intestinal malrotation into four types depending on the degree of rotation during the second stage of rotation: nonrotation, malrotation, reversed rotation, and paraduodenal hernia.

Intestinal malrotation can cause intestinal obstruction or midgut volvulus in infants. Approximately 64–80% of cases of intestinal malrotation present during the first few months of life [7]. The occurrence of intestinal malrotation in adults is rare because most patients remain asymptomatic. Thus, most cases of intestinal malrotation are incidentally found during abdominal examinations or operations. Moreover colon cancer with intestinal malrotation is very rare.

From 1974 to 2017 in Japan, 49 cases of colon cancer with intestinal malrotation, including our cases, were identified [8–17] (Table 1). However, a search of PubMed revealed seven cases of colon cancer with intestinal malrotation worldwide [11,18]. In the review of the literatures in Japan, the median patient age was 64 years (range, 22–88 years), and 27 patients were male. The tumors were located at the appendix in 1 case, cecum in 10 cases, ascending colon in 11 cases, transverse colon in 12 cases, sigmoid colon in 5 cases, descending colon in 2 cases and rectum in 9 cases. With respect to the type of intestinal malrotation, 34 (69.4%) cases were the nonrotation type, 8 (16.3%) cases were the reversed type, 5 (10.2%) cases were the malrotation type and 1 (2%) case was the paraduodenal hernia. Wang and Welch [5] reported that the malrotation type is the most common of the four types. Intestinal malrotation was diagnosed at surgery in 9 cases, by barium enema in 18 cases, by abdominal CT in 18 cases, by three-dimensional CT (3D-CT) angiography in 6 cases, by 3D-CT colonography in 3 cases, by virtual colonoscopy in 1 case, by multidetector-row CT in 1 case, and by sodium diatrizoate enema in 1 case. Although most cases of intestinal malrotation in adults were diagnosed by barium enema or during surgery until 2005, the development of imaging technology has increased the rate of diagnosis of intestinal malrotation by abdominal CT. At present, CT is one of the most useful diagnostic

modalities for intestinal malrotation in adults. Nehra and Goldstein [19] also reported that the diagnostic modality for intestinal malrotation was switched to abdominal CT from upper gastrointestinal series. Conversely, the tumor location is mostly diagnosed by barium enema and colonoscopy.

With respect to the surgical approach, open surgery was performed in 34 of 49 cases; laparoscopic surgery was performed in 15 (30.6%) cases. Until 2012, laparoscopic surgery was performed in only 2 of 27 (7.4%) cases. After 2012, however, laparoscopic surgery was performed 13 (59.1%) of 22 cases. This tendency is because laparoscopic surgery for colon cancer is becoming more widely performed, and the quality of this procedure is advancing. Although conventional laparoscopic colorectal cancer surgery with mesenteric excision is frequently and safely performed in the abdominal cavity, mesenteric excision outside the abdominal cavity was performed in 8 (53.3%) of 15 cases in a review of the literatures. Because intestinal malrotation is associated with abnormalities of the artery [16,20], it is difficult to safely perform laparoscopic lymph node dissection inside the abdominal cavity. A thorough preoperative understanding of the anatomical anomalies is important, especially vascular anomalies, for safe performance of mesenteric excision inside the abdominal cavity. In our review, three of four cases of malrotation diagnosed by 3D-CT angiography involved lymph node dissection inside the abdominal cavity. Therefore we think 3D-CT angiography is a useful modality for safe laparoscopic surgery in patients with colon cancer with intestinal malrotation.

#### 4. Conclusion

The laparoscopic approach for colon cancer with intestinal malrotation has not been established. The present review of the Japanese literature clearly showed that laparoscopic lymphadenectomy for colon cancer with intestinal malrotation is not feasible. If the intestinal malrotation is diagnosed preoperatively, 3D-CT

**Table 1**

Review of literature of intestinal malrotation with colon cancer in Japan except for the cases complicated with situs inversus totalis.

Case	Author	Publish	Age	Sex	Location	Type	Diagnosis of Intestinal Malrotation	Diagnosis of Tumor Location	Operation	Mesenteric Excision	Histopathology	Staging(UICC <sup>7th</sup> )
1	Hiratsuka	1974	47	F	cecum	nonrotation	operation	operation	open		unknown	unknown
2	Shimanuki	1988	73	M	rectum	nonrotation	barium enema	barium enema	open	tub1	unknown	unknown
3	Oshita	1993	68	M	rectum	nonrotation	barium enema	colonoscopy	open	tub2	T3,N2,M1a,StageIVA	
4	Isogai	1995	77	F	rectum	nonrotation	barium enema	barium enema	open	tub2	T2,N0,M0,StageI	
5	Yokota	1995	66	M	rectum	nonrotation	barium enema	operation	open	tub2	T4b,N1,M0,StageIIIC	
6	Ogawa	1997	69	F	transverse colon	unknown	barium enema	unknown	open	unknown	unknown	unknown
7	Sounaka	1997	22	M	ascending colon	nonrotation	operation	operation	open	tub1	T4a,N1,M0,StageIIB	
8	Kunieda	1998	57	F	rectum	nonrotation	barium enema	barium enema	open	asc	T2,N0,M0,StageI	
9	Kunieda	1998	62	F	rectum	malrotation	barium enema	barium enema	open	tub2	T4a,N0,M0,StageIIB	
10	Nagata	1998	72	M	rectum	reversed rotation	barium enema	colonoscopy	open	tub2	T3,Nx,M0,unknown	
11	Tamura	1999	55	M	cecum	nonrotation	barium enema	barium enema	open	por	T4a,Nx,M1b(P),StageIVB	
12	Sato	2001	60	M	appendix	nonrotation	barium enema	barium enema	open	muc	T2,N0,M0,StageI	
13	Sasaki	2003	71	F	cecum	malrotation	operation	colonoscopy	open	tub2	T3,N1,M0,StageIIB	
14	Fujita	2004	55	F	sigmoid colon	nonrotation	barium enema	colonoscopy	open	tub2	T3,N0,M0,StageIIA	
15	Uchida	2004	57	M	transverse colon	nonrotation	sodium diatrizoate	colonoscopy,selective arteriography	open	tub1	T3,N0,M0,StageIIA	
16	Oku	2005	56	M	ascending colon	nonrotation	barium enema	barium enema	open	tub1	M1a(H),StageIVA	
17	Tomimatsu	2005	81	F	ascending colon	nonrotation	barium enema	colonoscopy	open	tub1	T3,N2,M1a(H),StageIVA	
18	Sakaiwaza	2007	84	M	transverse colon	nonrotation	abdominal CT	abdominal CT	open	tub1	T4b,N1,M0,StageIIIC	
19	Yamamoto	2007	63	F	rectum	nonrotation	barium enema, MDCT	colonoscopy	laparoscopic	outside body	tub1	T2,N0,M0,StageI
20	Seki	2008	88	F	transverse colon	nonrotation	operation	colonoscopy	open	tub1	T3,N0,M0,StageIIA	
21	Nakasone	2009	71	F	sigmoid colon	nonrotation	abdominal CT	abdominal CT	open	tub1	T3,N1,M0,StageIIB	
22	Kobayashi	2009	60	M	ascending colon	reversed rotation	barium enema	colonoscopy	open	tub2	T3,N1,M0,StageIIB	
23	Itanari	2009	61	M	transverse colon	nonrotation	abdominal CT	abdominal CT	open	tub2	T3,N1,M0,StageIIB	
24	Takahashi	2009	84	M	ascending colon	nonrotation	barium enema	colonoscopy	laparoscopic	inside body	tub2	T3,N0,M0,StageIIA
25	Ito	2010	67	F	transverse colon	nonrotation	virtual colonoscopy	colonoscopy	open	tub1	T3,N2,M0,StageIIB	
26	Fukuhara	2010	76	F	cecum	nonrotation	operation	colonoscopy	open	tub1	T3,N1,M0,StageIIB	
27	Kokubo	2011	73	M	cecum	reversed rotation	abdominal CT	colonoscopy	open	tub2	T4b,N1,M0,StageIIIC	
28	Taiyoh	2012	53	F	sigmoid colon	nonrotation	abdominal CT	colonoscopy	open	tub2	unknown	
29	Sekizawa	2012	56	F	rectum	reversed rotation	abdominal CT	colonoscopy	open	tub2	T3,N2b,M0,StageIIIC	
30	Tokai	2012	79	M	transverse colon	nonrotation	3D-CT angiography	colonoscopy	laparoscopic	outside body	tub1	Tis,N0,M0,Stage0
31	Morimoto	2012	57	M	cecum	reversed rotation	abdominal CT	abdominal CT	open	unknown	T2,N0,M0,StageI	
32	our case	2013	78	M	cecum	nonrotation	operation	laparoscopic	outside body	tub1	T3,N0,M0,StageIIA	
33	Maeda	2013	48	M	transverse colon	nonrotation	3D-CT colonography	3D-CT colonography	open	tub2	T3,N0,M0,StageIIA	
34	Hirano	2013	82	F	transverse colon	reversed rotation	abdominal CT, barium enema	colonoscopy	laparoscopic	outside body	tub1	T1,N0,M0,StageI
35	Hirano	2013	68	F	ascending colon	malrotation	abdominal CT, barium enema	colonoscopy	laparoscopic	outside body	tub1	Tis,N0,M0,Stage0
36	Takahashi	2014	53	F	ascending colon	malrotation	abdominal CT, 3D-CT colonography	barium enema	laparoscopic	outside body	tub2	T3,N1,M0,StageIIB
37	Fujii	2014	73	F	cecum	nonrotation	abdominal CT	barium enema	open	tub1	Tis,N0,M0,Stage0	
38	Enomoto	2014	48	M	transverse colon	nonrotation	abdominal CT	barium enema	laparoscopic	inside body	tub1	T3,N0,M0,StageIIA
39	Kuroda	2014	64	F	transverse colon	nonrotation	abdominal CT	barium enema	laparoscopic	inside body	por	T4a,N1,M0,StageIIB
40	Morioka	2015	65	M	cecum	nonrotation	3D-CT angiography	colonoscopy	laparoscopic	inside body	tub2	T3,N0,M0,StageIIA
41	Kuwahara	2015	54	F	transverse colon	nonrotation	abdominal CT, 3D-CT angiography	colonoscopy	laparoscopic	inside body	tub2	T3,N0,M0,StageIIA
42	Kubota	2015	82	M	ascending colon	malrotation	abdominal CT,barium enema	colonoscopy	open	tub1	T4a,N2b,M0,StageIIIC	
43	Oshiro	2016	75	M	ascending colon	reversed rotation	abdominal CT,3D-CT angiography	colonoscopy,barium enema	open	tub1	T3,N0,M0,StageIIA	
44	Shima	2016	77	M	descending colon	reversed rotation	3D-CT angiography,3D-CTcolonography	colonoscopy,barium enema	laparoscopic	inside body	tub2	T3,N1,M0,StageIIB
45	Nakayama	2016	63	M	descending colon	nonrotation	abdominal CT	colonoscopy	open	tub2	T4b,N0,M0,StageIIC	
46	Motoki	2016	66	M	ascending colon	nonrotation	operation	colonoscopy	laparoscopic	outside body	tub2	T2,N0,M0,StageI
47	Nishida	2017	53	M	sigmoid colon	nonrotation	abdominal CT	colonoscopy	laparoscopic	inside body	tub1	T1,N0,M0,StageI
48	Kimura	2017	54	F	ascending colon	paraduodenal hernia	3D-CT angiography,abdominal CT	barium enema,double-balloon enteroscopy colonoscopy	open	tub2	T3,N0,M0,StageIIA	
49	our case	2017	81	M	cecum	nonrotation	operation	laparoscopic	outside body	tub1	Tis,N0,M0,Stage0	

tub1: well-differentiated tubular adenocarcinoma, tub2: moderately differentiated tubular adenocarcinoma, por: poorly differentiated adenocarcinoma, asc: adenosquamous carcinoma, UICC: Union for International Cancer Control, MDCT: multidetector-row computed tomography.

angiography should be used to reveal the vascular anatomic anomalies for safe performance of laparoscopic surgery.

## Conflicts of interest

The authors declare that they have no competing interests.

## Funding

None.

## Ethical approval

This paper was not a research study, so ethical approval not required.

## Consent

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

## Author contribution

KT made substantial contribution to conception and drafted the manuscript. KN conducted a literature search and made the contribution for acquisition of data. KT, KN, TS, KY performed the operation. KT, KN, KY and MK reviewed the manuscript and gave final approval for publication. KT was revising it critically for important intellectual content. All authors read and approved the final manuscript.

## Guarantor

The Guarantors of this manuscript are Katsushi Tokuhara and Prof. Masanori Kon.

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