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【 CASE REPORT 】

An Unusual Small Bowel Phytobezoar Successfully Resolved by Double-balloon Enteroscopy

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Abstract:
We herein report a very unusual case of small bowel obstruction caused by phytobezoar in a 69-year-old woman who consumed a large amount of bracken. The patient presented with nausea and vomiting. Computed tomography revealed an air-filled foreign body in the jejunum that had likely caused the small bowel obstruction. A fibrous foreign body diagnosed as a phytobezoar was detected using double-balloon enteroscopy. The obstruction was successfully resolved by crushing the phytobezoar repeatedly using a snare. Small bowel obstructions caused by phytobezoars are often treated with surgical interventions. However, endoscopic fragmentation using a snare is a minimally invasive treatment alternative.

Key words: small bowel obstruction, foreign body, small intestine, bracken, snare fragmentation

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Introduction

Bezoars are masses of indigestible foreign material. Usually found in the stomach, bezoars very rarely cause small bowel obstructions. Small bowel bezoars can clog the narrowest part of the small bowel at the ileocecal valve or the small bowel located 50-75 cm proximal to the ileocecal valve (1). Small bowel bezoars are difficult to diagnose and are usually surgically treated.

The development of double-balloon enteroscopy (DBE) in 2001 (2) made active endoscopic observation and endoscopic treatment of the entire small bowel feasible. However, reports of endoscopic treatment of small bowel bezoars are rare, with only five cases reported to date (1, 3-6).

We herein report the first case of small bowel obstruction caused by a phytobezoar due to over-consumption of bracken. The phytobezoar was treated endoscopically. In addition, a review of endoscopy treatment of small bowel bezoars is presented.

Case Report

A 69-year-old woman was referred to our hospital for nausea and vomiting. The patient had consumed a large amount of marinated bracken in soy sauce (Fig. 1a, b) four days before visiting the hospital. She had eaten the bracken five to six times per day, filling both hands at once. The patient had schizoaffective disorder, constipation, rheumatoid arthritis, and Sjögren's syndrome and was being treated with venlafaxine, levomepromazine, diazepam, lubiprostone, prednisolone, methotrexate, and tacrolimus. The patient had a history of total right hip replacement and no history of abdominal surgery. She had no fever, and her abdomen was mildly distended, soft, and not tender.

Laboratory findings included elevated white blood cell count and serum C-reactive protein (CRP); serum creatine kinase and lactate dehydrogenase were within normal limits (Table 1). Abdominal X-ray revealed a dilated small intestine (Fig. 1c), and contrast-enhanced computed tomography (CT) revealed an air-filled foreign body in the jejunum, with

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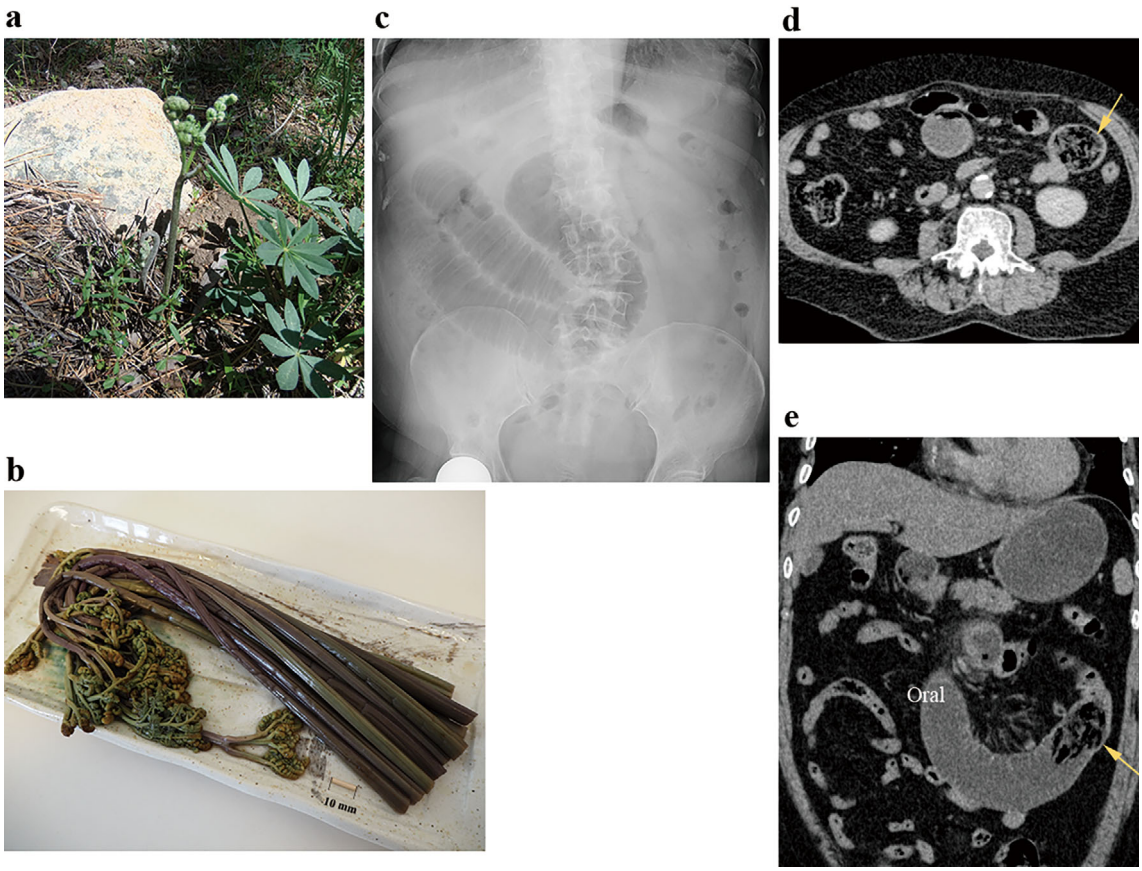

Figure 1. **a Bracken (*Pteridium aquilinum*), also known as eagle fern or *Warabi*, is a species of fern in the family Dennstaedtiaceae. Bracken is a wild vegetable that grows in grasslands, valleys, and the wilderness. In early spring, bracken produces fist-shaped sprouts that are edible. Bracken is widely eaten in Japan, China, and Korea. After removing the strong lye with baking soda, bracken is eaten as a boiled vegetable, pickled, marinated, or as an ingredient in miso soup. b Marinated bracken in soy sauce. c Abdominal X-ray showed a dilated small intestine. d, e Contrast-enhanced computed tomography revealed an air-filled foreign body (arrow) in the jejunum, with dilatation and fluid collection in the oral intestine.**

dilatation and fluid collection in the oral intestine (Fig. 1d, e). The patient's condition was diagnosed as a foreign body impaction in the small bowel.

As no intestinal ischemia was detected, nasogastric tube drainage was performed, followed by waiting for spontaneous evacuation. The inflammatory findings improved (CRP 0.5×10^{4} μg/L), and defecation occurred. However, contrast-enhanced CT indicated the presence of a foreign body in the small bowel causing incomplete impaction. After consulting surgeons, the foreign body was removed via DBE. Antegrade DBE revealed a 50-mm large, elastic hard, and dark greenish-yellowish foreign body in the jejunum with a shallow ulcer in the surrounding mucosa (Fig. 2a-c). The foreign body was strangulated with a snare and retracted directly into the instrument channel outlet, to crush the foreign body (Fig. 3a-d). After fragmenting the foreign body by repeated crushing, the endoscope was able to be advanced anally (Fig. 3e). Endoscopy-guided radiography showed the disappearance of the large filling defect and good passage through the intestinal tract (Fig. 3f). A cross-section of the fibrous foreign body suggested that it may have originated from ingested bracken, so the foreign body was ultimately diagnosed as a phytobezoar.

After treatment, abdominal X-ray and CT confirmed that the ileus had resolved, and the phytobezoar had disappeared (Fig. 4a-c). Oral intake was started two days after endoscopic treatment, and no recurrence of ileus was observed.

Discussion

There are two types of enteroliths: primary and secondary (7). Primary enteroliths are formed in the gastrointestinal (GI) tract and are further classified into "true" and "false" subtypes. True primary enteroliths are composed of substances found in chyme under normal gastrointestinal conditions, and they contain bile acids in the jejunum and calcium salts in the ileum, with bile acids stones having a calcified profile. These enteroliths are rare and are caused by a pathological condition in which intestinal contents are mechanically retained. False or pseudo-primary enteroliths are formed from insoluble foreign substances in the bowel due to the following three reasons: agglutination of a large

Table 1. Laboratory Findings and Evaluation of Salivary and Lacrimal Gland Functions.

Test	Patient's result	Reference range
White blood cell count	14.85×10^9 /L	3.50-8.90
Red blood cell count	4.6×10^{12} /L	3.78-4.97
Hemoglobin	136 g/L	112-149
Hematocrit	0.415 L/L	0.336-0.446
Platelet count	336×10^9 /L	125-375
Total protein	87 g/L	67-83
Albumin	41 g/L	40-50
Total bilirubin	11.97 μmol/L	3.42-22.23
Aspartate aminotransferase	26 IU/L	13-33
Alanine aminotransferase	37 IU/L	8-42
Alkaline phosphatase	236 IU/L	115-359
Lactate dehydrogenase	242 IU/L	119-229
γ-glutamyl transpeptide	25 IU/L	10-47
Creatine kinase	75 IU/L	45-163
Blood urea nitrogen	3.213 mmol/L	2.856-7.854
Creatinine	46.85 μmol/L	41.548-69.836
Na	138 mmol/L	138-146
K	3.9 mmol/L	3.6-4.9
Cl	99 mmol/L	99-109
C-reactive protein	13.04×10^4 μg/L	0.00-0.30
Rheumatoid factor	76 kU/L	0-15
ANA	1:320 (speckled)	<1:40
Anti-CCP antibodies	53.3 kU/L	<4.5
Anti-Ro/ SSA antibodies	1:64	Negative
Anti-La/ SSB antibodies	1:2	Negative
Sialography	Stage II[a]	Stage 0[a]
Chewing gum test	0 mL/10 min	>10
Schirmer test	Lt 6 mm/ Rt 5 mm	≥10
Fluorescent dye test	Positive	Negative

ANA: anti-nuclear autoantibodies, CCP: cyclic citrullinated peptide, Lt: left, Rt: right

[a]According to the criteria of Rubin and Holt (10).

amount of indigestible material (bezoar), precipitation of substances in the GI tract that become insoluble due to resorption of their solvents, and concentration of water-suspended insoluble salts and their mixture. Secondary enteroliths form in the extra-GI tract and migrate into the GI tract, causing obstruction, with gallstones being the most common type. Because no component analysis of the enteroliths was performed in this case, the biochemical characteristics of the enteroliths could not be determined. However, since no mechanical obstruction or extra-GI stones were noted, no calcification profile was detected in the jejunal enteroliths, and the clinical course suggested a vegetative bezoar, it was considered more likely to be a pseudo-primary enterolith like a phytobezoar rather than a true or secondary enterolith.

Mechanical small bowel obstruction in adults is an emergency condition that usually requires hospitalization and surgical intervention (8). Small bowel obstruction is usually caused by band adhesions or common hernias and is very rarely caused by foreign bodies or bezoars (8). The clinical symptoms of small bowel obstruction of bezoars include nausea, vomiting, abdominal pain, a fever, and an elevated white blood cell count (1, 5). Bezoars are aggregated poorly digested fibers of fruits and vegetables in the gastrointestinal tract (1, 5). Bracken, called *Warabi*, is a widely eaten wild vegetable in Japan. In the present case, the phytobezoar was caused by the peculiar eating behavior of the patient; the patient ate a large amount of marinated *Warabi* over a short period. This behavior may have been influenced by her schizoaffective disorder. In addition, reduced saliva secretion due to Sjögren's syndrome may have contributed to the development of the phytobezoar.

According to this patient's medical record, she was being treated with 48 μg/day lubiprostone by a psychiatrist and did not seem to have any problems with defecation; however, the detailed frequency of bowel movements could not be clarified retrospectively. Although the large bowel is generally the main site of fecal retention in constipation, and phytobezoar is rarely observed in the jejunum of patients with constipation, the possibility that constipation influenced this phytobezoar formation through mechanisms such as bowel dysfunction or microbiome disruption cannot be ruled out.

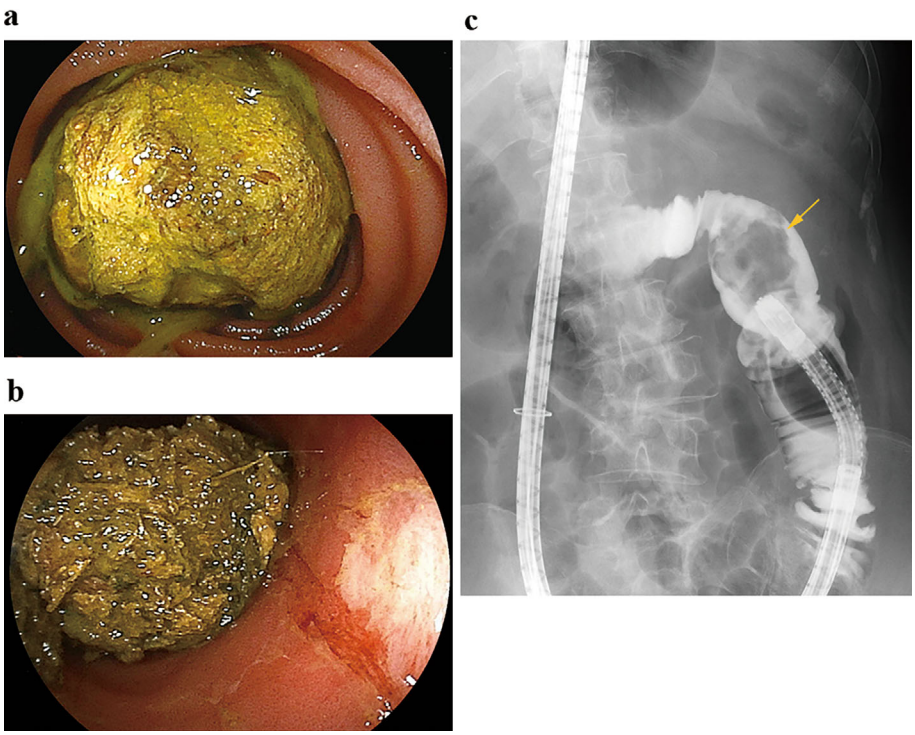

Figure 2. **a, b An irregular translucent mass (arrow) of approximately 50 mm in diameter in the jejunum, as shown using antegrade double-balloon enteroscopy (EN-580T; Fujifilm, Tokyo, Japan). c A shallow ulcer in the mucosa surrounding the foreign body.**

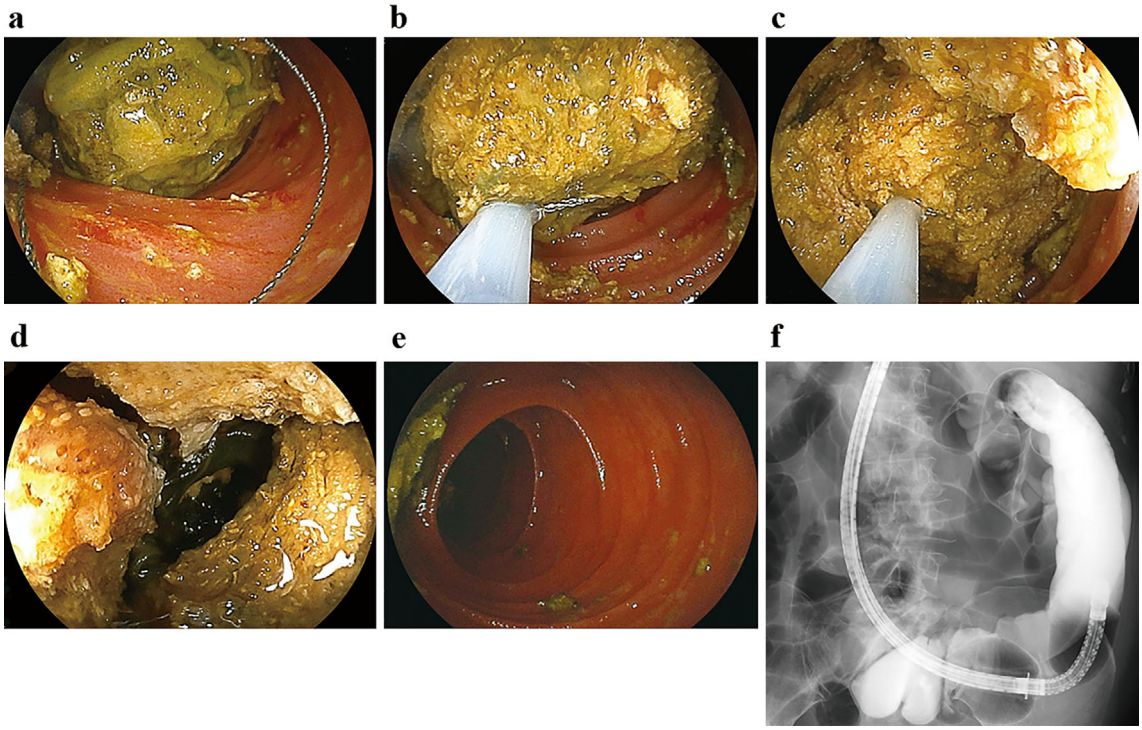

Figure 3. **a The foreign body was grasped with a snare (SD-210 U-25; Olympus, Tokyo, Japan), strangulated (b), and strongly pulled directly into the endoscope forceps channel outlet of the endoscope for shredding (c, d). Repeating this procedure multiple times crushed the foreign body, and the endoscope was able to be advanced toward the anus (e). After crushing, the large translucent image disappeared, and good passage through the intestinal tract was confirmed by enteroscopy-guided radiography using gastrografin (f).**

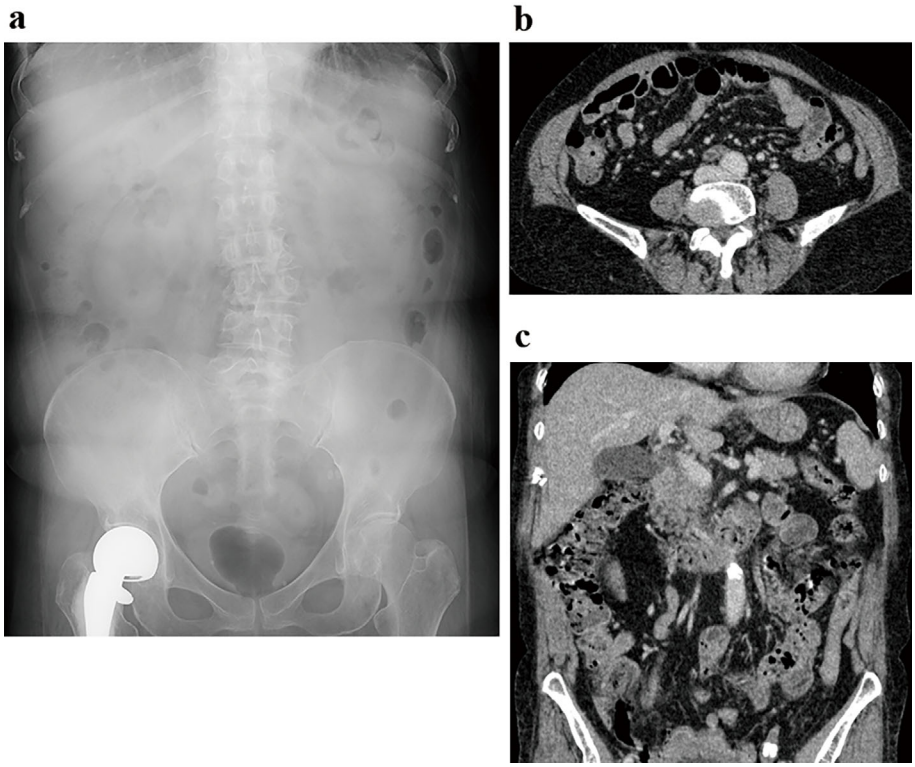

Figure 4. **No findings of ileus or a residual foreign body were noted on abdominal X-ray (a) and CT images (b, c) after enteroscopy treatment.**

Table 2. **Clinical Characteristics of the Present and Previous Reports of Endoscopic Treatment of Small Bowel Bezoars.**

Age/ sex	Background	Symptom	Bezoars					Enteroscopy, devices	Endoscopic treatment	Outcome
			Location	Source	Size (mm)	CT findings	Endoscopic findings			
70/F (1)	HT, T2DM, c/o	Abdominal pain, nausea, and vomiting	Distal jejunum	Agar	50	Masses outlined by fluid	White-colored, hard	DBE, forceps, and a snare	Unsuccessful fragmentation	Extraction with enterotomy
68/F (3)	A pyloroplasty and a hysterectomy	Epigastric pain and vomiting	Proximal jejunum	Fragrant manjack	>50	Ovoid-shaped mass with mottled gas	Green mass	DBE, a snare, and a Dormia basket	Successful fragmentation	No recurrence
62/M (4)	DM	Intermittent, cramping abdominal pain	Ileum	ND	40-50	Fecal-like mass with air bubbles	Whitish round mass along with ulcers	SBE, forceps, and a snare	Successful fragmentation	Surgery[a]
69/M (5)	ND	Epigastralgia	Distal ileum	Persimmons	40	Mass with air contents	Yellow, hard	DBE	Unsuccessful	Extraction with enterotomy
80/F (6)	HT	Abdominal pain and vomiting	Jejunum	Tannin-containing foods[b]	50	Mass with air bubbles	A yellowish, impacted foreign body	DBE	ND	Segmental resection
69/F (the present case)	Schizoaffective disorder, RA, SS	Nausea and vomiting	Jejunum	Marinated bracken	50	Air-filled foreign body	Dark greenish-yellowish foreign body with ulcers	DBE, a snare	Successful fragmentation	No recurrence

F: female, M: male, HT: hypertension, T2DM: type 2 diabetes mellitus, c/o: cesarean operation, DBE: double-balloon enteroscopy, SBE: single-balloon enteroscopy, ND: not described, RA: rheumatoid arthritis, SS: Sjögren's syndrome

[a]One week after the fragmentation using enteroscopy, the patient had abdominal pain again, and surgery was performed to remove the bezoar from the ileum.

[b]Analysis of the biopsy specimen revealed that 98% of the bezoar was composed of tannin acid, and she was diagnosed with small bowel obstruction by migration of a gastric bezoar. Thus, diospyrobezoar, a subtype of phytobezoars caused by persimmons, was suspected.

CT is useful for diagnosing small bowel obstructions, such as the presence and localization of phytobezoars (9). The common CT findings for phytobezoars include a round or ovoid mass containing mottled gas at the obstructed site. The mass may have an encapsulating wall. In the present case, the phytobezoar was an air-filled foreign body, as shown by CT. The five previously reported cases of phytobezoars showed similar findings (Table 2). Thus, a mottled air-filled mass in the small bowel should raise suspicion of phytobezoar.

In the management of small bowel obstruction caused by phytobezoar, DBE is both a powerful diagnostic tool and a treatment method. Six case reports describe attempts to treat phytobezoar via endoscopy, including the present case (Table 2) (1, 3-6). Macroscopically, phytobezoars are white to yellow-green masses that occupy the small bowel lumen and may cause ulceration of the surrounding mucosa. Only two cases of phytobezoars were completely treated via endoscopy: the present case and a case in which a snare and a Dormia basket were used to fragment the phytobezoar (3). In one case (4), the phytobezoar was fragmented, but bowel obstruction recurred, leading to surgery. The phytobezoar should be crushed as finely as possible, and snare impaction during the fragmentation should be avoided. In the present case, the phytobezoar was safely treated by grinding the edges. Furthermore, the phytobezoar was difficult to fragment only by strangulation with a snare; crushing the phytobezoar by forcefully pulling it into the endoscope forceps channel outlet after strangulation was extremely effective.

In conclusion, detailed patient interviews concerning eating behavior and unique CT findings are essential for the diagnosis of small bowel obstructions caused by phytobezoars. While surgical treatment is common, careful endoscopic fragmentation using a snare is an effective therapeutic option for treating phytobezoars.

The authors state that they have no Conflict of Interest (COI).

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