

[CASE REPORT]

Phytobezoar Associated with Levodopa-carbidopa Intestinal Gel Infusion in Patients with Parkinson's Disease: A Case Report and Literature Review

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Abstract:

Continuous intrajejunal infusion of levodopa-carbidopa intestinal gel (LCIG) is an established device-aided therapy for advanced Parkinson's disease (PD). Phytobezoar associated with LCIG is a rare device-related complication and presents with exacerbations of gastrointestinal and PD symptoms. We herein report the case of a phytobezoar that was formed at a knot on the pigtail-shaped J-tube and developed only in association with postprandial abdominal pain, similar to a feeling of a tube being pulled in without an exacerbation of PD symptoms. Such abdominal pain may be a warning sign of phytobezoar in LCIG-treated patients. Despite device-related complications, high-pressure alarms are not always present, and PD symptoms are not always exacerbated.

Key words: levodopa-carbidopa intestinal gel, parkinson's disease, phytobezoar, device-aided therapy, J-tube, gastrointestinal symptom

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Introduction

Continuous intrajejunal infusion of levodopa-carbidopa intestinal gel (LCIG) is an established device-aided therapy for patients with advanced Parkinson's disease (PD) suffering from motor fluctuations and dyskinesia (1, 2). As LCIG infusion is conducted by performing gastrostomy, insertion of a percutaneous endoscopic gastrostomy (PEG)-J tube, and use of a portable pump, procedure/device-associated adverse events (AEs) have become problematic (1-3). AEs are associated with PEG-J tube occlusion/dislocation, accidental removal of PEG-J tube, and pump malfunction (1-3).

Bezoars are indigestible conglomerations trapped in the gastrointestinal tract that are named according to their composition. The most common type of bezoar is the phytobezoar, which consists of undigested fiber, fruit seeds, or vegetables (4). The risk factors for bezoar formation include a

history of gastric surgery, ingestion of fiber-rich foods, and suffering from systemic diseases, such as hypothyroidism and diabetes mellitus (4). Phytobezoar associated with LCIG is a rare device-related complication that forms compressive erosion, ulcerative lesions, and fistulas due to tube extension in the gastrointestinal tract and can be fatal at times (5-13).

We herein report a case of phytobezoar that was formed at a knot on the pigtail-shaped PEG-J tube and developed only with abdominal symptoms, without the exacerbation of PD symptoms and pump malfunction. We also provide a literature review of phytobezoars associated with LCIG.

Case Report

A 56-year-old Japanese man had been suffering from PD since 2009. He underwent PEG in 2018 due to motor fluctuation and dyskinesia, and the continuous infusion of LCIG was initiated. In December 2019, the J-tube was changed

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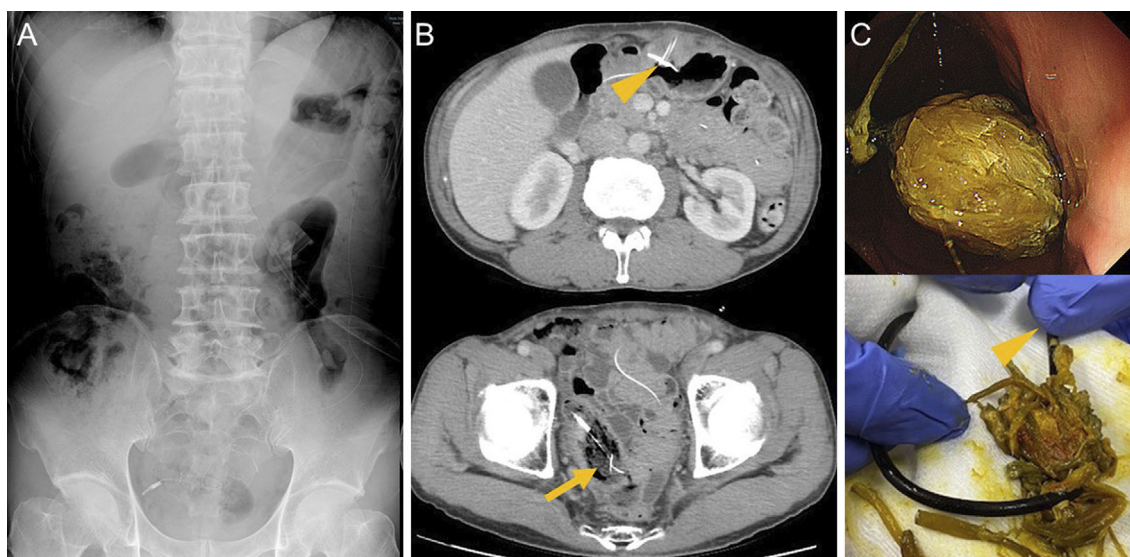


Figure. (A) Abdominal radiography shows that the phytobezoar is lodged deep in the pelvic jejunum. (B) Computed tomography shows a hypodense mass containing air in the jejunum at the tip of the J-tube (arrow), and the position of the gastric fixation plate is maintained (arrowhead). (C) Upper gastrointestinal endoscopy shows an entangled undigested vegetable at the knot at the pigtail-shaped of the J-tube, forming a phytobezoar (diameter: 2.5 cm). The arrowhead indicates the tip of the J-tube.

because it was occluded due to kinking and knotting. In September 2020, he noticed that the PEG tube had become inserted quite deeply after the external fixation plate of the PEG-J tube had come off on awakening. Following this episode, he visited our department because of postprandial abdominal pain. Postprandial abdominal pain appeared within minutes of eating and lasted for two to three hours, similar to the feeling of a PEG-J tube being pulled in. However, his neurological symptoms were stable, and the LCIG pump alarm detected no high pressure. His medical history was unremarkable, and he had no history of gastrointestinal tract surgery or taking laxatives.

A physical examination showed slight abdominal tenderness at the right side of the PEG, no abdominal distension, and no rebound abdominal tenderness. The Movement Disorder Society-Sponsored Revision of the Unified Parkinson's Disease Rating Scale (MDS-UPDRS) Part III was 11 points during the "on" time. The LCIG infusion parameters were as follows: rate 2.8 mL/h, morning dose 16 mL, and extra dose 2.2 mL. The J-tube was able to be flushed. Abdominal radiography showed that the tip of the J-tube had been inserted deep into the pelvic jejunum, and a knot was found at the tip of the J-tube. (Figure A). Abdominal computed tomography displayed a hypodense mass containing air in the jejunum at the tip of the J-tube, and the position of the gastric fixation plate was maintained (Figure B).

Because phytobezoar of the tip of the J-tube was suspected, the J-tube was manually removed under X-ray fluoroscopy and endoscopy. Upper gastrointestinal endoscopy showed that an undigested vegetable had gotten entangled at the knot at the pigtail-shaped part of the J-tube, forming a phytobezoar (Figure C). After removing the J-tube, the

postprandial abdominal pain disappeared. One week later, J-tube replacement was performed, and the gastrointestinal and neurological symptoms have remained stable since.

Discussion

We described a case of phytobezoar associated with LCIG infusion in a patient with advanced PD that caused postprandial abdominal pain without PD symptom exacerbation.

Continuous intrajejunal infusion of LCIG in patients with advanced PD is useful for controlling motor complications; however, device-related complications mainly manifest as AEs (2). Despite PEG-J tube occlusion due to kinking/knotting, PEG-J tube dislocation has been the most frequent device-related complaint (2, 3).

Phytobezoar is a rare complication of LCIG that sometimes induces compressive erosion, ulcerative lesions, and fistulas due to tube extension in the gastrointestinal tract and requires surgical intervention (5-13). Phytobezoars associated with LCIG are caused by dietary fiber-rich foods and intestinal hypomotility as a non-motor symptom of PD (5, 6, 11). The clinical features of patients with phytobezoars associated with LCIG are presented in Table and include (i) a period between J-tube insertion and the development of symptoms due to phytobezoar ranging from seven weeks to four years; (ii) symptoms of phytobezoar associated with LCIG commonly including gastrointestinal symptoms, such as abdominal pain and/or worsening of PD symptoms; (iii) pump malfunctions not always occurring; (iv) the J-tube potentially not being easily flushed; and (v) endoscopy often showing an ulcer due to compression of the

Table. Clinical Features of Patients with Phytobezoar Associated with LCIG.

Reference	Age/ Sex	Period from the J-tube insertion	Abdominal symptom	PD symptom	Pump	Flushing	Radiography	CT: Detection of hypodense mass	Endoscopy: Detection of ulcer
5	71/M	5 m	Pain	NR	High pressure alarm	Not be flushed	NR	NR	Erosion of the gastric mucosa
5	69/M	7 w	Pain	NR	Normal	Easy	NR	NR	Multiple pyloric and jejunal ulcers
6	74/M	23 m	Nausea	Not affected	Normal	NR	Passing through the duodenal wall of the tube	NR	Erosion of the gastric mucosa
7	74/M	1 y	Pain	Dysphasia of solids	NR	NR	NR	NR	Large antro- pyloric ulcer
8	70/M	4 y	None	Motor deterioration	NR	NR	NR	NR	None
11	68/F	2 y	Pain	NR	NR	NR	NR	NR	Ulcerated bedsores
9	58/F	2.5 y	NR	Worsening of motor symptoms	NR	NR	NR	NR	Duodenal ulcer
10	70/M	13 m	Pain	Worsening of motor symptoms	NR	NR	Dislocation of the jejunal tube in the small bowel	NR	Duodenal ulcer
12	42/F	429 d	Pain	NR	NR	NR	NR	NR	Ulcer
13	55/F	2 y	Pain	NR	NR	NR	NR	NR	Duodenal ulcer
Present case	56/M	9 m	Pain	Not affected	Normal	Easy	Dislocation of the J-tube in the pelvic jejunum	A hypodense mass at the tip of the J-tube	None

LCIG: levodopa-carbidopa intestinal gel, F: female, M: male, NR: not reported, d: days, w: weeks, m: months, y: years, PD: Parkinson's disease

J-tube.

In the present case, the patient complained of postprandial abdominal pain without exacerbation of PD symptoms and malfunction of the infusion pump nine months after the J-tube had been inserted. Upper gastrointestinal endoscopy showed no ulcers. Ulcerative lesions of the jejunum could not be evaluated endoscopically in this case. As there were no ulcerative lesions, we hypothesized that the postprandial abdominal pain resulted from traction of the J-tube with the phytobezoar moving to the anal side due to intestinal peristalsis. It is important to be aware of the fact that a phytobezoar can cause abdominal pain, even in the absence of ulcerations. Furthermore, the J-tube had a knot; however, there were no abnormalities in flushing. As a result, there was likely no high-pressure alarm on the pump. Therefore, it should be noted that only some abdominal pain was experienced by the patient, which was a feeling similar to that of a PEG-J tube being pulled in without loss of clinical efficacy or malfunction of the infusion pump in cases of phytobezoar associated with LCIG.

In the present case, a phytobezoar was suspected based on the findings of the knot at the tip of the J-tube and a hypodense mass containing air at that site on CT. In general, phytobezoars are visualized on CT as ovoid or round masses in the gastrointestinal tract with air bubbles retained inside

and a mottled appearance (14). However, CT can also miss these features, as the densities of an intestinal phytobezoar and ordinary intestinal stool mass are similar (15). Imaging studies have also revealed intestinal distension, dislocation of the PEG-J tube, and enteroenteric fistula (5, 9-11, 13). Abdominal CT might be useful for confirming the fistula, intestinal obstruction, and phytobezoar.

The management of a phytobezoar associated with LCIG includes endoscopic removal, surgical extraction, and dissolution by the administration of Coca-Cola[®] (5-13). Furthermore, it is important to avoid fiber-rich foods, such as asparagus, burdock, butterbur, bracken, leek, and bean sprouts. Therefore, when ingesting these foods, patients should cook them by chopping them into small pieces and boiling them sufficiently in order to soften them and thereafter chew them thoroughly (5). In the present case, the PEG-J tube was removed using X-ray fluoroscopy and endoscopy. The phytobezoar contained leek and bean sprouts, and the food had not been properly chewed. The dietary fiber was considered to have been part of the standard intake in this case, but it is probable that inadequate chewing, chopping, and cooking were inadequate as well. As the phytobezoar was formed with the J-tube knot as the core, we speculated that the entanglement of food residues in the J-tube knot caused the phytobezoar. When patients with PD use LCIG, adequate

dietary guidance must be provided. Dietary guidance was provided to this patient after the removal of the phytobezoar to help prevent recurrence.

In conclusion, the occurrence of abdominal pain associated with a feeling that the PEG-J tube is being pulled in may be a warning sign of a phytobezoar in patients treated with LCIG. Abdominal CT may be useful for detecting phytobezoars; therefore, it is important to consider the presence of a phytobezoar and interpret this accordingly. Furthermore, the possibility of the presence of a phytobezoar should always be considered if the patient exhibits abdominal pain, as there is not always an exacerbation of high-pressure alarms or PD symptoms.

The patient provided his written informed consent for the publication of this report. Procedures were conducted in accordance with the Declaration of Helsinki.

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

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