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Case Report

Successfully Treated Pneumatosis Cystoides Intestinalis with Pneumoperitoneum Onset in a Patient Administered α -glucosidase Inhibitor

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An 80-year-old woman, who had been administered α -glucosidase inhibitor for diabetes, was brought to the hospital with the sensation of abdominal fullness and pain. Abdominal computed tomography indicated pneumatosis cystoides intestinalis (PCI) in the small intestinal wall, with free air within the abdomen. A blood examination showed no increases in white blood cells or C-reactive protein level. The patient's condition improved with conservative therapy. PCI with pneumoperitoneum induced by α -glucosidase inhibitor is rare, with only 27 cases (excluding the present case) reported in Japan to date. In PCI with pneumoperitoneum, differentiation from gastrointestinal perforation is important and following the clinical symptoms over time is vital.

Key words: pneumatosis cystoides intestinalis, pneumoperitoneum, α -glucosidase inhibitor

Pneumatosis cystoides intestinalis (PCI) is a relatively rare disorder in which multilocular or linear air cysts form in the subserosa or submucosa of the intestinal tract wall [1]. The cause of PCI is unknown, but the case of some individuals with this disorder that may have been induced by alpha (α)-glucosidase inhibitors have been reported in recent years [2-30]. We encountered a patient who had been administered α -glucosidase inhibitor and developed PCI with pneumoperitoneum. This case was successfully cured with conservative treatment, and we report our findings herein.

Case Report

An 80-year-old woman complained of feelings of abdominal bloating and abdominal pain while at an elder daycare facility and visited her previous doctor. Plain radiography of the abdomen showed free air, and since peritonitis was therefore suspected, she was brought to our hospital by ambulance. She had no history of heavy alcohol intake or smoking, and no family history of note. She had undergone cholecystectomy for cholelithiasis at 38 years of age. At 71 years of age, she underwent hysterectomy for uterine cancer and received radiotherapy as adjuvant therapy. At 58 years of age, she became paralyzed on the right side of the body following a cerebral infarction. She was taking an α -glucosidase inhibitor for diabetes. There were no complications relevant to diabetes in this case. In addition, she was routinely taking mag-

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nesium oxide and purgatives for chronic constipation, along with oral medications such as antihypertensive agents. When she was admitted to our hospital, she had abdominal bloating and subcutaneous emphysema. Only mild tenderness was observed on palpation of the abdomen, with no muscular guarding or rebound tenderness. The results of physical examination on admission were as follows: body height, 138 cm; body weight, 43 kg; blood pressure, 204/91 mmHg; body temperature, 36.8°C; heart rate, 69 beats/min; respiratory rate, 18 breaths/min; and peripheral oxygen saturation, 95%. The patient underwent blood testing and various imaging modalities. No findings indicative of marked inflammatory reactions were observed from the results of complete blood count or blood biochemistry (Table 1). Abdominal radiography showed distension of the intestine and an extraintestinal gas image, suggesting pneumoperitoneum (Fig. 1). Abdominal computed tomography (CT) revealed intraperitoneal free air and a gas image in the intestinal tract wall (Fig. 2A–C).

Based on the above findings, PCI with pneumoperitoneum was diagnosed. Concerning the pathological conditions that present with intraperitoneal free

air, many types of perforations were considered as differential diagnoses, including perforation of gastrointestinal ulcerations or tumors, perforation of a diverticulum of the intestine, and gastrointestinal perforation triggered by radiotherapy for uterine cancer. However, based on the CT findings, PCI was considered most likely. Conservative treatment was selected, because of the minor increase in inflammatory reaction and mild abdominal pain. The patient was immediately hospitalized and monitoring was started with the administration of transfusion and antibiotics while the patient fasted. We used cefmetazole sodium for this patient. The administration of α -glucosidase inhibitor was discontinued. Her general status remained stable and the symptom of abdominal bloating became less severe on the day after hospitalization, along with the passage of flatus. On Day 2 after hospitalization, another abdominal CT was conducted, showing decreased gas (Fig. 2D). Improvement of PCI was confirmed and led us to prohibit water intake for the patient. Because her symptoms were stable following water intake, dietary intake was started on Day 4. No exacerbation of abdominal

Table 1 Blood biochemical findings. No findings indicating marked inflammatory reactions were observed from complete blood count or biochemical examination

WBC	6,010	/ μ l	CK	79	mg/dl
RBC	434	10,000/ μ l	UN	18	mg/dl
Hb	13.7	g/dl	Cr	0.5	mg/dl
Ht	41.2	%	UA	3	mg/dl
PLT	25	10,000/ μ l	CRP	0.06	mg/dl
TP	7.1	g/dl	Na	136	mEq/l
Alb	4.2	g/dl	K	4.1	mEq/l
T-Bil	0.44	mg/dl	Cl	104	mEq/l
AST	29	IU/l	FBS	108	mg/dl
ALT	18	IU/l	PT-INR	0.9	
γ GTP	19	IU/l	APTT	28.3	Sec
LDH	279	IU/l	D dimer	6.3	μ g/ml

WBC, white blood cell; RBC, red blood cell count; Hb, hemoglobin; Ht, hematocrit; PLT, platelet; TP, total protein; Alb, albumin; T-Bil, total bilirubin; AST, aspartate amino transferase; ALT, alanine aminotransferase; γ GTP, gamma glutamyl transpeptidase; LDH, lactic acid dehydrogenase; CK, creatine phosphokinase; UN, blood urea nitrogen; Cr, creatinine; UA, uric acid; CRP, C reactive protein; Na, Sodium; K, potassium; Cl, chlorine; FBS, fasting blood sugar; PT-INR, prothrombin time international normalized ratio; APTT, activated partial thromboplastin time.



Fig. 1 Abdominal X-ray showing distension of the intestine and an extraintestinal gas image under suspicion of pneumoperitoneum.

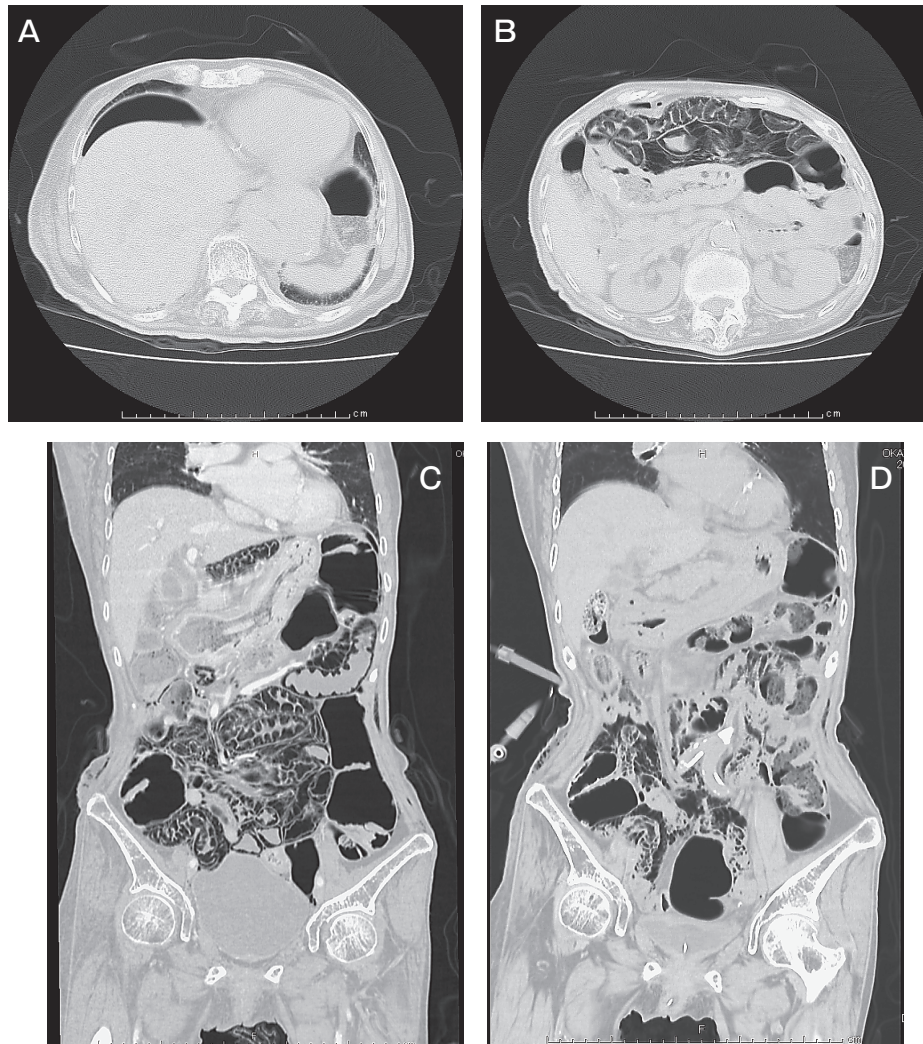


Fig. 2 Abdominal CT. **A**, Intraperitoneal free air is apparent on the surface of the liver in the upper abdominal area; **B, C**, Gas image in the intestinal tract wall, mainly in the small intestine; **D**, On CT conducted 2 days after hospitalization, the gas image shows a decreasing tendency.

symptoms was observed thereafter and she was moved to the referring hospital on Day 6.

Discussion

With PCI, multiple air cysts are formed in the submucosa or subserosa of the intestinal tract wall. Abdominal pain, constipation, and a feeling of abdominal bloating are representative symptoms. Various causes for the development of PCI have been considered, and the following theories have been reported. The first option is that intestinal inner pressure increases, causing intestinal gas to invade the

inside of the intestinal tract wall through a damaged area of mucosa [31]. Second, gas-producing bacteria such as *Clostridium* may pass through the intestinal tract wall and produce gas in the intestinal tract wall [32]. Third, air leaks may occur through alveoli damaged due to a lung disorder, reaching the intestinal membrane or intestinal tract wall via the mediastinum/retroperitoneum, and forming air bubbles [33]. Fourth, the condition may develop due to exposure to chemicals such as trichloroethylene [34]. In addition, in cases of collagen disease or with long-term administration of steroids, a weakened intestinal tract wall may be the cause [4, 9, 10, 12, 22, 23]. However,

in recent years, some cases probably induced by α -glucosidase inhibitors, as oral diabetes drugs, have been reported. These α -glucosidase inhibitors act to limit increases in blood sugar levels by inhibiting the degradation of disaccharide into saccharide, thus inhibiting and delaying the absorption of glucide. With this action, glucide is not completely digested and reaches the inside of the large intestine from the lower small intestine. Through this mechanism, incompletely digested glucide remains present and promotes increases in intestinal bacteria and fragmentation by enzymes of these bacteria, yielding organic acids such as lactic acid and butyric acid, along with intestinal

gases such as hydrogen gas and methane gas. Due to the increased presence of gas in the intestine, side effects of α -glucosidase inhibitors include abdominal bloating and flatulence. Taniyama *et al.* suggested that the associated increase in internal pressure in the intestine contributes to PCI [35]. Excluding the present patient, 27 case reports of PCI induced by α -glucosidase inhibitors in Japan and 2 cases in other country were identified using the keywords “pneumatosis cystoids intestinalis” and “ α -glucosidase inhibitor” or “pneumatosis cystoids intestinalis” and “diabetes mellitus” in a search of the Japan Centra Evuo Medicina and Pubmed (Table 2). The range of age of

Table 2 Reported cases of *Pneumatosis cystoides intestinalis* in patients internally administered α -glucosidase inhibitor

	Age	Gender	Major complaints	Surgery	Outcome	Person who reported the case/year
1	87	Female	Abdominal bloating	×	Remission	Azami [2] 2000
2	73	Female	Abdominal bloating	×	Remission	Tachibana [3] 2002
3	55	Female	Abdominal bloating	×	Remission	Maeda [4] 2002
4	73	Female	Abdominal pain	Peritoneal washing, drainage	Remission	Matsuda [5] 2004
5	unknown	Female	Abdominal pain	×	Remission	Furio [6] 2006
6	65	Male	Abdominal pain/Diarrhea	×	Remission	Miyakawa [7] 2006
7	66	Male	Abdominal bloating	×	Remission	Nagahara [8] 2006
8	56	Female	Abdominal bloating	×	Remission	Hisamoto [9] 2006
9	77	Female	Abdominal pain	Exploratory laparotomy	Remission	Maeda [10] 2007
10	75	Male	Abdominal bloating	×	Remission	Yasuoka [11] 2007
11	69	Male	Abdominal pain	×	Remission	Tsujimoto [12] 2008
12	58	Male	Nothing in particular	×	Remission	Hosoi [13] 2008
13	79	Female	Abdominal bloating	×	Remission	Kinuta [14] 2008
14	65	Female	Abdominal pain	×	Remission	Vogel [15] 2008
15	71	Female	Abdominal pain/Vomiting	Exploratory laparotomy	Remission	Gondo [16] 2009
16	73	Female	Abdominal bloating/Abdominal pain	Exploratory laparotomy	Remission	Kuwada [17] 2010
17	60	Male	Abdominal bloating/Abdominal pain	×	Remission	Kusano [18] 2010
18	88	Male	Abdominal bloating	Exploratory laparotomy	Remission	Kasumoto [19] 2010
19	58	Male	Abdominal pain	×	Remission	Kojima [20] 2010
20	91	Male	Abdominal pain/Vomiting	×	Remission	Misuta [21] 2010
21	48	Male	Nothing in particular	×	Remission	Shimajima [22] 2011
22	68	Male	Nothing in particular	×	Remission	Igata [23] 2011
23	89	Female	Abdominal pain	Exploratory laparoscopy	Remission	Matsuda [24] 2011
24	67	Female	Abdominal pain	Exploratory laparotomy	Remission	Ishioka [25] 2011
25	66	Male	Nothing in particular	×	Remission	Nakaji [26] 2011
26	80	Female	Abdominal bloating	×	Remission	Shigetoshi [27] 2011
27	70	Male	Abdominal bloating	×	Remission	Matsuura [28] 2011
28	76	Male	Abdominal pain	×	Remission	Hashimoto [29] 2011
29	67	Male	Abdominal bloating/Abdominal pain	×	Remission	Miyamoto [30] 2012
30	80	Female	Abdominal bloating/Abdominal pain	×	Remission	Our case 2013

the patients was 48 to 91 years old (average, 70.6 years). Three patients had a renal disease, 2 patients had a collagen disease, 2 patients had a pemphigus, and 1 patient had a myasthenia gravis. Six of the patients had received long-term steroid treatment. Among the above theories regarding PCI development, the administration of α -glucosidase inhibitors applies to the present case. In addition, internal pressure in the abdomen would have been increased physiologically due to chronic constipation and decreased vermuculation from diabetes. As treatment, conservative management can be used for PCI if the internal administration of α -glucosidase inhibitor is discontinued and symptoms recover with fasting. Oxygen therapy is also reportedly effective [5, 6, 8, 11, 17, 23, 26, 28]. In terms of the underlying mechanism, gas in the intestinal tract wall in PCI is normally thought to be nitrogen gas, and the administered oxygen generates a denitrogenation action [36]. However, in cases with pneumoperitoneum like the present case, differential diagnosis from gastrointestinal perforation is important, as mentioned above. When clinical symptoms progress, operative therapy should be considered and the patient should be monitored carefully.

In conclusion, We encountered a successfully treated case of PCI with pneumoperitoneum. An α -glucosidase inhibitor was considered one of the main pathogenic factors in this case of PCI. In PCI with pneumoperitoneum, differentiation from gastrointestinal perforation is quite important, and the strict follow-up of clinical symptoms over time is vital.

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