Case Report

Spontaneous Gastric Perforation in an Adolescent with Severe Motor and Intellectual Disabilities: A Case Report

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Summary

Spontaneous gastric perforations are rare in adolescents. Here, we report the case of a 15-year-old boy with severe motor and intellectual disabilities (SMID) who was diagnosed with spontaneous gastric perforation. Due to his original condition and poor peristalsis caused by antiepileptic drugs, gas formation in the stomach and intestines was severe since infancy. After a femoral fracture at the age of 14 years and 5 months, the patient experienced reduced food intake and weight. He transferred to our hospital with shock vital due to severe peritonitis caused by gastric perforation. Immediately, he underwent an emergency laparotomy and partially resected with margins at the site of gastric perforation at the age of 15 years and 1 month. The cause of the gastric perforation was not identified by pathology. Intraoperative ascites culture detected *Candida albicans*. He was administered a long-term antifungal treatment for enteric mycosis with high β-D-glucan levels. Two months after the surgery, he was discharged. The detailed mechanism of gastric perforation is unclear but increased lumen pressure in the gastrointestinal tract has generally been speculated. In our case, we considered that the gastric perforation was caused by intestinal immunocompromise with *Candida albicans* overgrowth, associated with increased lumen pressure owing to the transverse position of the stomach. Therefore, deflation using gastrostomy may have been effective in relieving excess luminal pressure. Adequate nutritional evaluation and gastrostomy should be considered in patients with SMID, who have massive intestinal gas formation and gastric bubbles.

Key Words: *Candida albicans*, Gastric perforation, Malnutrition, Pediatric surgery, Severe motor and intellectual disabilities

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Introduction

Gastric perforations in children are usually associated with trauma, peptic ulcers, and drug use. Therefore, spontaneous gastric perforation of an unknown cause rarely occurs during childhood, especially in adolescence[1]. Although there are various theories on the occurrence of spontaneous gastric perforation, it remains unexplained. We report an adolescent case of spontaneous gastric perforation in a 15-year-old boy with severe motor and intellectual disabilities (SMID).

The patient was diagnosed with cerebral palsy and a congenital brain malformation. During adolescence, his spinal scoliosis worsened, and weight loss was observed due to inadequate oral intake and malnutrition. During surgery for spontaneous gastric perforation, ascitic fluid culture revealed the presence of *Candida albicans*. Herein, we review the patient's clinical course and discuss the mechanism of spontaneous gastric perforation during adolescence.

Case Presentation

The patient was born at 37 weeks of gestation via spontaneous delivery. The motor developmental milestones were delayed in the early infantile stage, and the child was diagnosed with severe motor and intellectual disabilities (SMID). Based on magnetic resonance imaging, the patient was diagnosed with cerebellar dysplasia and cerebral cortical dysplasia. At the age of 14 years, he was 143 cm tall and weighed 25.0 kg. Because he had difficulty swallowing, his parents spent a great deal of time feeding him. We recommended gastrostomy; however, they denied to gastrostomy because they felt feeding him as rewarding. The patient also had recurrent aspiration pneumonia. Nutritional products were not used because of the increased risk of aspiration. Due to his original condition and poor peristalsis caused by antiepileptic drugs, he had excessive gas formation in the stomach and intestines since infancy. He also had osteoporosis and had fractured the right femur at 14 years and 5 months of age. After this event, the patient's weight decreased to 22.5 kg. Blood tests at that time did not reveal malnutrition (Table 1). Thereafter, his oral intake decreased from 14 years and 11 months of age.

At the age of 15 years and 1 month, the patient presented with lethargy and shock. The abdomen was distended and skin redness was observed. In addition, he had abdominal muscle rigidity, which is a clear sign of peritonitis. Blood examination showed that aspartate aminotransferase, alanine aminotransferase, lactate dehydrogenase, γ-glutamyl transferase levels were 15, 7, 194, and 19 U/L, respectively. There was no increase in total bilirubin levels. Total protein and albumin levels were 8.3 and 4.7 g/dL respectively. Probably due to dehydration, urea nitrogen levels were elevated at 29.2 mg/dL. Creatinine level was reduced to 0.3 mg/dL due to his thinness and low muscle volume. Electrolytes such as sodium, potassium, and chloride were within normal ranges. The C-reactive protein and procalcitonin levels were 0.3 mg/dL and 0.05 ng/mL, respectively. Peripheral blood test showed an elevated white blood cell count of 19.2 10^9/L. There was no anemia, low platelet count, or abnormal coagulation data (Table 1). The blood culture results were negative. A nasogastric tube was inserted and 500 mL dark red fluid was drained. Radiography revealed free air in the abdomen and severe scoliosis (Fig. 1A). Enhanced computed tomography revealed free air on the ventral side of the stomach, reticulum, right Morison's pouch, and porta hepatis (Fig. 1B). However, the perforation site could not be identified. Subsequently, the patient's blood pressure decreased to 75/40 mmHg, and treatment with catecholamines was initiated.

We performed emergency laparotomy based on the diagnosis of gastrointestinal perforation. Laparotomy was performed through an upper midline incision, and a 10 mm diameter perforation was found on the lesser anterior wall of the upper stomach (Fig. 2). For pathological diagnosis and evaluation of blood flow in the dark lesion around the perforation site, we performed partial gastric resection with a 10 mm margin and sutured it there after checking for bleeding from the resected edge. We collected contaminated ascitic fluid for culture and washed the abdominal cavity with 5 L of normal saline. Subsequently, the abdominal cavity was irrigated, four drains were placed, and the wound was closed. The patient was admitted to the intensive care unit (ICU) postoperatively. Pathologically, the perforated hole measured 10 × 5 mm, the muscularis propria at the perforated edge was bluntly ruptured, and the mucosa and muscularis propria around the perfora-

Table 1 Results of venous blood tests 8 months before (at the age of 14 years and 5 months) and at the time of hospital visit (at the age of 15 years and 1 month)

Venous blood test	Results 8 months before hospital visit	Results at the time of hospital visit	Unit
Aspartate aminotransferase	13	15	U/L
Alanine aminotransferase	7	7	U/L
Lactate dehydrogenase	141	194	U/L
Total bilirubin	0.3	0.4	mg/dL
Total protein	7.1	8.3	g/dL
Albumin	3.8	4.7	g/dL
Urea nitrogen	5.0	29.2	mg/dL
Creatinine	0.23	0.30	mg/dL
Na	140	134	mmol/L
K	4.1	3.3	mmol/L
Cl	109	101	mmol/L
Creatine kinase	117	23	U/L
Procalcitonin	NA	0.05	ng/mL
C-reactive protein	0.53	0.29	mg/dL
pH	7.35	7.19	
pCO_2	39.4	55.4	mmHg
pO_2	54.0	36.1	mmHg
Base excess	−3.9	−8.0	mmol/L
HCO_3	21.3	20.8	mmol/L
SvO_2	87.7	57.0	%
Lactic acid	1.6	4.2	mmol/L
White blood cell	4.9	19.2	10^9/L
Neutrophil	2.6	16.8	10^9/L
Red blood cell	4.8	5.3	10^12/L
Hemoglobin	13.6	15.1	g/dL
Platelet	28.2	29.2	10^4/dL
Prothrombin time		14.2	sec
Prothrombin Time INR		1.20	
Activated partial thromboplastin time		36.4	sec

Na; sodium, K; potassium, Cl; chloride, pCO_2; partial pressure of carbon dioxide, pO_2; partial pressure of oxygen, HCO_3; bicarbonate, SvO_2; mixed venous oxygen saturation

tion site showed a high degree of neutrophil aggregation. However, the pathology concluded that the cause could not be identified (Fig. 3 and 4). Consequently, the patient was diagnosed with idiopathic gastric perforation. On the third postoperative day, his breathing had improved from ventilator support to spontaneous breathing. On the following day, the patient was discharged from the ICU. At that time, his weight was 18.5 kg. Despite the administration of piperacillin/tazobactam, he had a prolonged fever persisted. *Candida albicans* was cultured from an intraoperative culture of the ascitic fluid, but periodic acid-Schiff (PAS) and Grocott staining of surgical specimens did not confirm the presence of *Candida albicans*. His blood β-D glucan level was high at 126.0 pg/mL. He was treated with fluconazole for 2 months, followed by which his prolonged fever improved.

After initiating tube feeding and rehabilitation, the patient was discharged at the age of 15 years and 3 months. He underwent gastrostomy at 16 years of age. Since then, his status stabilized, and he gained weight.

Discussion

Spontaneous gastric perforation is often idiopathic and not caused by factors such as trauma, peptic ulcers, and drug administration; however, it has no clear definition. In neonates, gastric wall ischemia due to neurogenic disorders, vascular shunts, or diving reflexes can cause spontaneous gastric perforation[2, 3]. In preschool children, increased intraluminal pressure

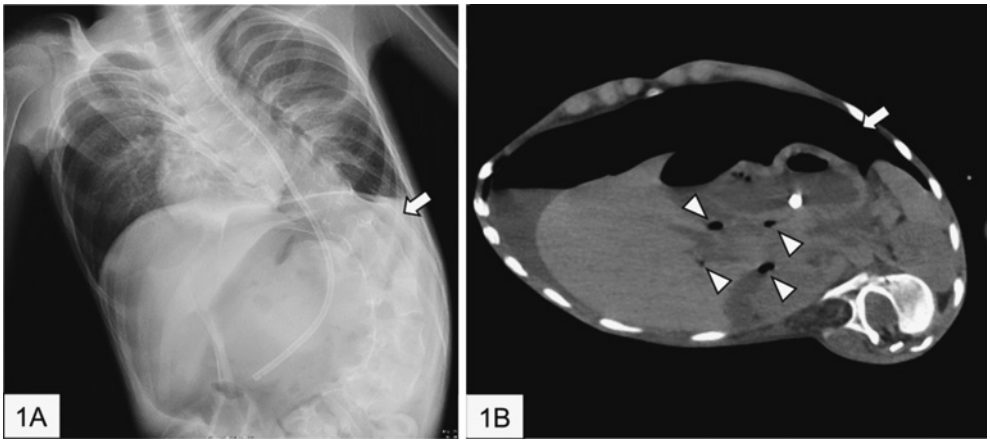

Figure 1 Abdominal radiograph and computed tomography image
A: Abdominal radiograph shows intra-abdominal free air (arrow) and severe scoliosis. **B:** Enhanced computed tomography image reveals free air at the ventral side of the stomach (arrow), reticulum, right Morison's pouch, and porta hepatis (arrowheads).

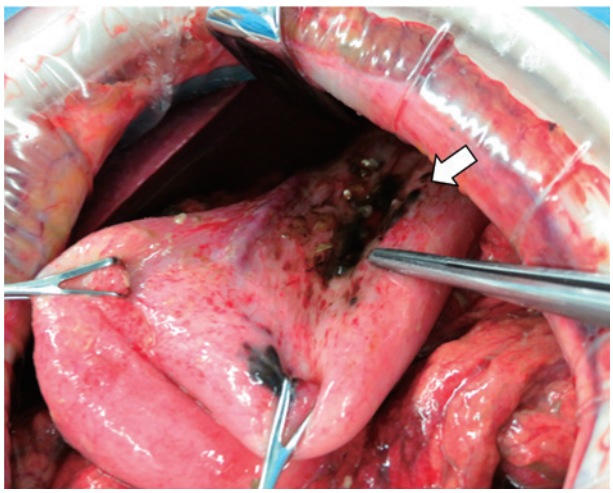

Figure 2 Intraoperative photograph
Intraoperative photograph shows 10-mm diameter perforation (arrow) on the lesser anterior wall of the upper stomach.

may cause spontaneous gastric perforation[4, 5]. Additionally, gastric wall ischemia has been suggested to contribute to perforation[4]. In adults with spontaneous gastric perforations, ruptures often occur around the lesser curvature of the stomach. It has been proposed that this is because a small curvature has a low elastic modulus owing to the lack of mucosal folds and the small curvature is subjected to greater stretching than the rest of the stomach when it expands to form a spherical shape[3, 4]. It is more common in adult women, and spontaneous gastric rupture due to increased abdominal pressure has been reported in pregnant women. The proposed mechanism in these patients is that high intraluminal pressure causes impaired perfusion and/or obstruction of venous drainage, leading to necrosis of the gastric wall[5]. Our patient was small in stature but almost adult in age. As previously reported, the gastric perforation was on the lesser curvature of the stomach.

In the present case, *Candida albicans* was detected in the ascitic fluid culture. The question of whether this was the cause or the effect of the perforation was raised. Candida species are endemic to 30-60% of healthy people[6]. Gastric candidiasis has been reported in 0.96% of upper gastrointestinal endoscopy[7]. Candida species are controlled by beneficial bacteria and a low pH environment. Therefore, immunosuppression (e.g., diabetes, HIV infection, and chemotherapy) or suppression of gastric acid secretion by antacids can cause gastrointestinal candidiasis[8, 9]. The patient was severely dehydrated and in shock when he visited our hospital due to gastric perforation, and it is difficult to assess his nutritional status based on the data at that time. Although the patient had osteoporosis and was expected to be in poor nutritional condition originally, the most recent blood tests before the onset of the gastric perforation showed no malnutrition. Disappointingly, that data was collected eight months before the perforation occurred, even though his oral intake had markedly declined two months before it occurred. More extensive and frequent blood tests could have predicted the gastric perforation. However, there have been re-

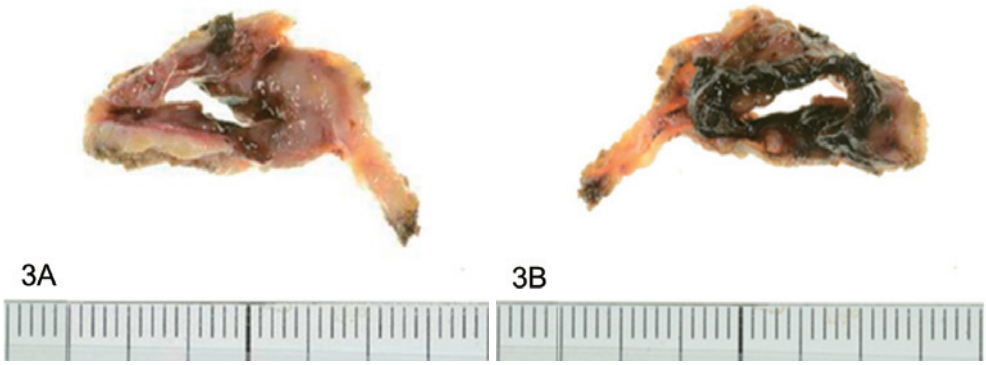

Figure 3 Photographs of the excised specimen
A: The mucosal side of excised specimen shows that the perforation is not ulcerated. **B:** The serosal side of excised specimen shows the black edge of the perforation site.

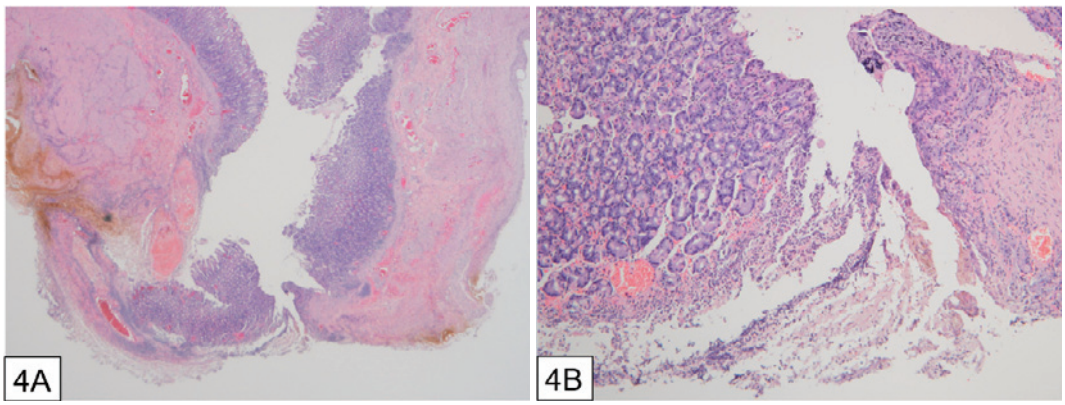

Figure 4 Histopathological assessment of the excised specimen
A: Hematoxylin-eosin staining (×20) shows a bluntly ruptured muscularis propria at the perforated edge. **B:** Hematoxylin-eosin staining (×100) shows a high degree of neutrophil aggregation in the mucosa and muscularis propria around the perforation site.

ports of gastric perforation by Candida species in healthy person[9]. In the present case, the stomach was originally distended as if it was about to perforate. Gastric perforation may have occurred because of Candida overgrowth caused by compromised immunity due to inadequate nutrition, combined with luminal hypertension due to the transverse position of the stomach. Deflation using gastrostomy may have been effective in relieving excess luminal pressure.

In gastric perforations caused by Candida species, the margins of the perforation may be black or gray[9, 10]. Perioperative specimens are more important than postoperative specimens for confirming gastrointestinal perforation caused by Candida species. The diagnosis of gastric perforation due to gastric candidiasis is made by demonstrating the presence of Candida in PAS- or Grocott-stained specimens[10, 11]. Although the margins of the perforation were black in the present case (Fig. 3B), the presence of Candida species was not demonstrated endoscopically or pathologically. Therefore, we could not determine whether gastrointestinal candidiasis was the cause of gastric perforation.

In patients with gastrointestinal perforation, the duration of ICU stay, ventilator use, and central venous catheter use were reported to be significantly longer in cases where Candida was detected in intraoperative ascitic fluid cultures than in cases where Candida was not detected[12]. If Candida species are detected in the

ascitic fluid or blood cultures in cases of gastric perforation, antifungal therapy for a minimum of 2 weeks is recommended[11]. In the present case, β-D glucan level was used to determine the efficacy of the treatment because blood cultures did not show Candida species, but it did not become negative for a long period. Fluconazole was administered for 2 months during prolonged hospitalization for nutritional management and rehabilitation, with no adverse effects.

We report a case of spontaneous gastric perforation and gastrointestinal candidiasis in a boy with SIMD. In patients with SMID and massive intestinal gas and gastric bubble formation, gastric perforation may occur when oral intake is reduced. Although emergency surgery and administration of antifungal medication saved the patient's life, this case reminds us that adequate nutritional evaluation and gastrostomy should be considered for such patients.

Informed Consent

Written informed consent was obtained from the patient's parents for the publication of this report and the associated images.

Author Contributions

Developing the concept of the report: S.W and K.S

Providing the figures: S.N and Y.N

Writing the manuscript: S.W

Editing the manuscript: S.M, T.Y, K.O, S.N, Y.N, Y.O, G.I, M.M, M.T, S.T, M.N, S.M, T.N, K.S, S.Y, and K.K

All authors approved the submission of the final article.

Disclosure Statement

None declared (there is no conflict of interest).

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