

CASE REPORT

A case of intussusception caused by cecal duplication after COVID-19

Rika Satoyoshi*, Kei Kotanagi, Takuya Kichiraku, Kei Yoshino, Kazuhiro Kudoh, Toshiya Sawada, Hideaki Miyazawa and Hitoshi Kotanagi

Department of Gastroenterological Surgery, Akita Red Cross Hospital, Akita, Japan

*Correspondence address. Department of Gastroenterological Surgery, Akita Red Cross Hospital, 222-1 Naeshirosawa-Saruta, Kamikitade, Akita 010-1495, Japan. Tel: +81-188295000; Fax: +81-188295255; E-mail: rika_satoyoshi@akita-med.jrc.or.jp

Abstract

Coronavirus disease 2019 (COVID-19) was thought to have respiratory symptoms as the main manifestation, but it has become clear that extrapulmonary symptoms such as gastrointestinal disorders also occur. There are several reports of intussusception associated with COVID-19 in children, but these are rare in adults. In this report, we present a case of cystic intestinal duplication that enlarged during the course of COVID-19 treatment and resulted in intussusception. Right hemicolectomy was performed for intussusception due to the cystic lesion. To the best of our knowledge, this is the first resected case of intussusception due to alimentary tract duplication after COVID-19 infection.

INTRODUCTION

The outbreak of severe acute respiratory syndrome-coronavirus-2 (SARS-CoV-2) that causes coronavirus disease 2019 (COVID-19) has a significant impact in the entire world. Although COVID-19 is first defined by its respiratory symptoms, it is now recognized that the virus can also affect the digestive system causing gastrointestinal symptoms like diarrhea, loss of appetite, nausea/vomiting and abdominal pain as a major complaint [1]. Intussusception is rare as a symptom of COVID-19, and most of the reports are pediatric cases. We report an adult, surgical case of intussusception caused by cystic alimentary tract duplication that enlarged right after the process of respiratory failure due to COVID-19.

CASE PRESENTATION

A 63-year-old male who had fever and dyspnea visited a medical institution. Both antigen and polymerase chain reaction (PCR) tests for SARS-CoV-2 were positive and he was hospitalized for treatment. Nine days after admission, he was transferred to our hospital due to deteriorating respiratory condition. After admission to our hospital, ventilator management and steroid administration were performed. After 3 days, his respiratory condition improved and he was able to withdraw from the

ventilator. Computed tomography (CT) taken for observation of lung disease coincidentally showed cystic lesions in the right abdomen (Fig. 1). He had no abdominal symptoms at this time. The patient was discharged on the 18th day of hospitalization. He had diarrhea from the 15th day after discharge and abdominal pain from the next day. He consulted his family doctor on the 17th day because the abdominal pain persisted. CT examination revealed enlargement of cystic lesion and intussusception of the right colon (Fig. 2). The patient was transferred to our hospital as an emergency case. Physical findings showed fever with a body temperature of 38°C and tenderness in the right lower abdomen. Blood test showed an inflammatory reaction with a white blood cell count of 13 000/ μ l and C-reactive protein of 2 mg/dl. The SARS-CoV-2 PCR testing was negative. We performed emergency surgery on the same day.

Intraoperative findings

A round mass existed in proximal colon that caused invagination. We performed right hemicolectomy.

Gross findings of excised specimens

A cystic lesion originated from cecum, with a necrotic discoloration of the cyst wall. When the cyst was incised,

Received: January 28, 2022. Accepted: February 14, 2022

Published by Oxford University Press and JSCR Publishing Ltd. All rights reserved. © The Author(s) 2022.

This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<https://creativecommons.org/licenses/by-nc/4.0/>), which permits non-commercial re-use, distribution, and reproduction in any medium, provided the original work is properly cited. For commercial re-use, please contact journals.permissions@oup.com

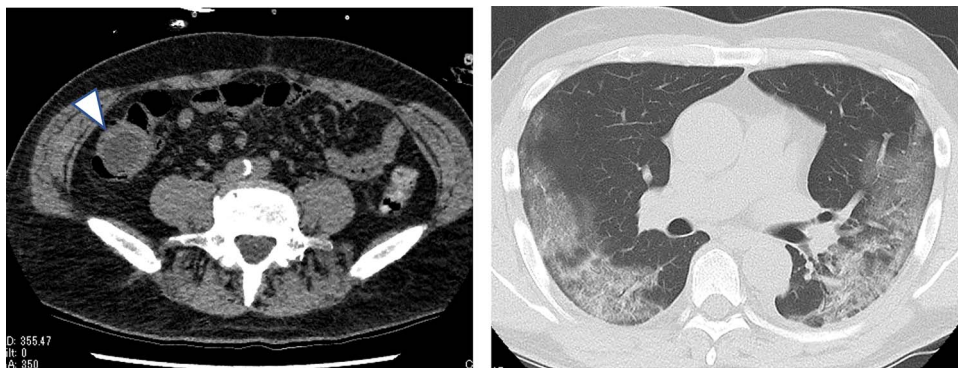


Figure 1. Plain CT on the 13th day after COVID-19 onset (the day after withdrawal from the ventilator); a cystic lesion was found in the right lower abdomen (arrowhead).

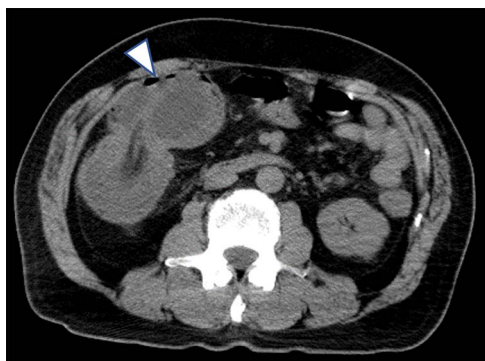


Figure 2. Plain CT taken when the patient was transferred to our hospital for abdominal pain; the cystic lesion had increased in size (arrowhead) and showed intussusception.

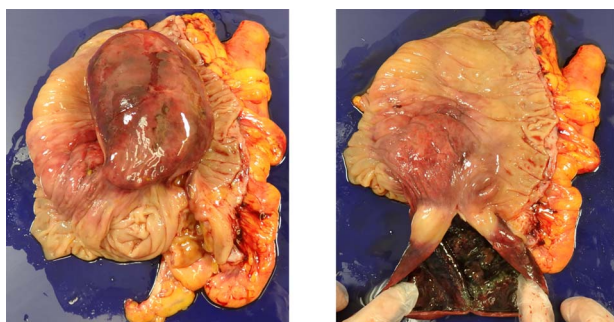


Figure 3. Excised specimen; there was a cystic lesion in the cecum, and the inner lumen of the cyst was necrotic.

it contained a brown malodorous liquid inside, and the inner surface of the cyst was also necrotic (Fig. 3).

Pathological findings

Both the inside and outside of the wall were covered with colonic mucosa, and there was a muscularis mucosae in the submucosa, but no muscularis propria. Erosion was prominent on the mucosa inside the cyst, and diffusely severe inflammatory cell infiltration and edema were observed under the mucosa. There was no malignancy. From these findings, the lesion was diagnosed as intussusception of colon due to cecum cystic duplication.

After the operation, there were no problems, including respiratory complications, and the patient was discharged 11 days after surgery.

DISCUSSION

Initially, the aspect of COVID-19 as a severe respiratory tract infection was emphasized, but now it is known as a disease that also causes systemic symptoms such as the central nervous, cardiovascular and digestive systems. The prevalence of gastrointestinal disease in COVID-19 has been reported to be as high as 40%, the most common of which were nausea or vomiting, diarrhea and anorexia [2]. The pathophysiological mechanisms relating these gastrointestinal symptoms to COVID-19 are not yet elucidated. Angiotensin-converting enzyme 2 (ACE2) receptor is the functional receptor for SARS-CoV-2. ACE2 is not exclusive to lung cells and is also present in cells of the gastrointestinal tract, specifically in the epithelial tissue of the esophagus, ileum and colon, which is considered to be one of the causes of injuring the gastrointestinal epithelium and causing gastrointestinal symptoms [3].

There have been several reported cases of intussusception associated with COVID-19 worldwide, but most are pediatric cases [2–9], and as far as we know, there is only one report of adult case [10]. In some of these reports, lymphoid hyperplasia secondary to acute viral infection is one of the causes of intussusception. In general, intussusception in adults is account for ~5% of all intussusceptions and the underlying cause of only 1% of bowel obstruction [11]. In adult cases, intussusception often has an organic disease such as a tumor. There are reports of cases of intussusception caused by intestinal duplication, but most are reported in young adults aged 19–32 [11–15]. Gastrointestinal tract duplications are uncommon congenital abnormalities that manifest before the age of 2 years in 80% of cases [15]. By definition, they are located in or adjacent to the wall of part of the gastrointestinal tract and have smooth muscle in their walls and are lined by alimentary tract mucosa. Both events are unusual especially in adults, and the combination of both is even more uncommon [15]. In our

case, the cystic lesion in cecum gradually increased in size in the process of COVID-19. This case had diarrhea early after discharge, and it is possible that COVID-19 caused inflammation of the intestinal tract. Inflammation of the intestinal tract may secondarily occlude an opening of the duplication cyst, causing an increase in cystic lesions and resulting in intussusception.

CONCLUSION

As COVID-19 continues around the world, there is a possibility that the number of cases with abdominal diseases requiring surgery will increase, and it is necessary to pay attention to gastrointestinal complications as well as respiratory disorders.

CONFLICT OF INTEREST STATEMENT

None declared.

FUNDING

None.

REFERENCES

1. Nabil A, Elshemy MM, Uto K, Soliman R, Hassan AA, Shiha G, et al. Coronavirus (SARS-CoV-2) in gastroenterology and its current epidemiological situation: an updated review until January 2021. *EXCLI J* 2021;**20**:366–85.
2. Jin B, Singh R, Ha SE, Zogg H, Park PJ, Ro S. Pathophysiological mechanisms underlying gastrointestinal symptoms in patients with COVID-19. *World J Gastroenterol* 2021;**27**:2341–52.
3. Guerrón N, Figueroa LM. Intussusception and COVID19, successful mechanic reduction, case report. *Glob Pediatr Health* 2021;**8**:1–3.
4. Cai X, Ma Y, Li S, Chen Y, Rong Z, Li W. Clinical characteristics of 5 COVID-19 cases with non-respiratory symptoms as the first manifestation in children. *Front Pediatr* 2020;**12**:258.
5. Moazzam Z, Salim A, Ashraf A, Jehan F, Arshad M. Intussusception in an infant as a manifestation of COVID-19. *J Pediatr Surg Case Rep* 2020;**59**:101533.
6. Mercado-Martínez I, Arreaga-Gutiérrez FJ, Pedraza-Peña AN. Intussusception and SARS-CoV-2 infection. *J Pediatr Surg Case Rep* 2021;**67**:101808.
7. Osorno JF, Giraldo M, Marín AF, Figueroa LM. Novel coronavirus infection in an infant with intussusception. *Glob Pediatr Health* 2021;**8**:1–3.
8. Giovanni JE, Hrapcak S, Melgar M, Godfred-Cato S. Global reports of intussusception in infants with SARS-CoV-2 infection. *Pediatr Infect Dis* 2021;**40**:e35–6.
9. Bazuaye-Ekwuyasi EA, Camacho AC, Rios FS, Torck A, Choi WJ, Aigbivbalu EE, et al. Intussusception in a child with COVID-19 in the USA. *Emerg Radial* 2020;**27**:761–4.
10. Jackson KM, Sabbota AL. Right hemicolectomy for ileocolonic intussusception in an adult with active COVID-19 infection: a case report. *J Surg Case Rep* 2021;**6**:1–3.
11. Al-Qahtani HH. Enteric duplication cysts as a leading point for ileoileal intussusception in an adult: a rare cause of complete small intestinal obstruction. *World J Gastrointest Surg* 2016;**8**:472–5.
12. Kim HS, Sung JY, Park WS, Kim YW. An ileal duplication cyst manifested as an ileocolic intussusception in an adult. *Turk J Gastroenterol* 2014;**25**:196–8.
13. Ho YC. Total colorectal and terminal ileal duplication presenting as intussusception and intestinal obstruction. *World J Gastrointest Surg* 2012;**18**:6338–40.
14. Kyo K, Azuma M, Okamoto K, Nishiyama M, Shimamura T, Maema A, et al. Laparoscopic resection of adult colon duplication causing intussusception. *World J Gastrointest Surg* 2016;**22**:2398–402.
15. Reiser-Erkan C, Erkan M, Ulbrich E, Nährig J, Kleeff J. Cystic colon duplication causing intussusception in a 25-year-old man: report of a case and review of the literature. *BMC Surg* 2010;**10**:19.