INTRODUCTION

The development of capsule endoscopy (CE) and balloon-assisted enteroscopy (BAE) has enabled endoscopists to manage obscure gastrointestinal bleeding (OGIB). Among patients with OGIB, Meckel’s diverticulum (MD) is an important differential diagnosis for OGIB. Although MD is usually asymptomatic, it can cause various clinical symptoms such as gastrointestinal (GI) bleeding, intestinal obstruction, and acute abdominal pain. The prevalence of Meckel’s diverticulum is approximately 0.3%–2.9% in adults. Of them, 4% develop related complications, and obstruction is considered a major manifestation of adult symptomatic MD. Although it varies across studies, the mean reported distance from the ileocecal valve is approximately 50 cm, where it is often difficult to reach using a conventional colonoscope. While there are several specific examinations used to diagnose MD, including CE, BAE using a single or double balloon, technetium-99m pertechnetate scan, and arteriography, it is occasionally
difficult to obtain a definite diagnosis of MD. Resection of MD is necessary when judged as a source of clinical manifestations.

CE is less invasive than BAE, and its diagnostic ability is superior to BAE for patients with OGIB. Therefore, CE is usually the first method used to identify OGIB. Although CE is considered to be a safe procedure, there is a risk of capsule retention. Capsule retention is defined as the capsule remaining in the GI tract for more than 2 weeks. The main cause of capsule retention is a small intestinal stricture. The reported etiology of small intestinal stricture includes drug-induced small intestinal stricture due to nonsteroidal anti-inflammatory drugs, tumors, inflammatory bowel disease, postoperative stricture, and stricture induced by radiation. Here, we report a case of simultaneous small intestinal bleeding and stricture due to MD.

2 | CASE REPORT

A 44-year-old man with a history of occasional melena since childhood presented to our hospital with a complaint of hematochezia. The patient had no irregular bowel habits. Physical examination revealed no abnormal findings. Laboratory evaluation revealed decreased hemoglobin levels (10.4 mg/dl). No bleeding source or dilation of the bowel was identified on abdominal contrast-enhanced computed tomography (CT). The patient underwent esophagogastroduodenoscopy and total colonoscopy. Although bright red blood was seen on colonoscopy, the source of bleeding was not identified. Furthermore, the source of bleeding was not detected on abdominal contrast-enhanced computed tomography (CT). The patient was diagnosed with OGIB, and CE was performed 10 days after colonoscopy to examine the small intestine. Normal intestinal fluid was observed by CE, and it was difficult to identify the source of bleeding. The CE was evacuated without any regional transit abnormality. We explained the necessity of performing BAE to the patient; however, he did not consent to BAE. Fortunately, the bleeding stopped spontaneously. Magnetic resonance imaging in the outpatient department at the follow-up did not identify the source of bleeding. The patient was prescribed rebamipide at the follow-up. After 9 months, the patient presented to our hospital with lower GI bleeding. Since detailed testing including CE had been performed with previous bleeding, BAE was the first test performed after obtaining informed consent. BAE revealed a diverticulum in the ileum at 50 cm from the ileocecal valve, and intestinal stricture was observed on the anal side of the diverticulum (Figure 1A). It was difficult to traverse the scope (SIF-260; Olympus Co., Tokyo, Japan), and the small bowel series with Gastrografin showed a blind end that corresponded to the diverticulum and the stricture (Figure 1B). Although no active bleeding was observed during the BAE procedure, there were no other possible sources of bleeding. Therefore, we diagnosed MD as the source of bleeding and decided to treat the patient surgically. The diverticulum was apparent, and segmental small bowel resection with primary anastomosis was performed (Figure 2A, B). Pathology of the surgical specimen showed a true diverticulum with a small erosion (Figure 2C), and heterotopic gastric mucosa was seen in the diverticulum; therefore, MD was diagnosed pathologically (Figure 2D). The patient had no lower GI bleeding since the surgery.

3 | DISCUSSION

We encountered a patient with OGIB who eventually experienced a small intestinal stricture due to MD. The clinical course of this patient suggests two important clinical issues. First, although the frequency is low, patients with OGIB may have small intestinal strictures due to inflammation from MD. Second, the diagnosis of bleeding due to MD is still difficult to determine, even when using several diagnostic modalities.

In the present case, intestinal stricture due to MD was diagnosed by BAE after the second occurrence of
hematochezia. Although capsule retention was avoided in this patient, transient capsule retention inside the MD has been reported in other cases.\textsuperscript{11} A possible explanation of the clinical course of the present case was that bleeding from the ulcer in the ileum occurred; thereafter, stricture developed as a result of scar formation after ulcer healing. The ulcer was formed due to exposure to acid secretion from the heterotopic gastric mucosa of MD. Because we performed CE after the second occurrence of hematochezia, there was a possibility that capsule retention might occur since the stricture was too severe to traverse with an enteroscope. More commonly, capsule retention due to drug-induced stricture from nonsteroidal anti-inflammatory drugs has been reported, usually presenting as diaphragmatic stricture.\textsuperscript{12,13} Rezapour et al. conducted a meta-analysis of retention associated with CE and reported that capsule retention occurred in 2.1\% of patients with OGIB.\textsuperscript{10} Furthermore, 3\% of patients with capsule retention in this meta-analysis developed obstructive symptoms. Patency CE is used for patients who have a high risk of intestinal stricture, and reports indicate that it reduces the risk of capsule retention by half in IBD patients.\textsuperscript{10} Performing patency CE for all OGIB patients is not recommended, considering the low incidence of stricture due to MD (such as in our case), but endoscopists should keep in mind the possibility of stricture due to MD when examining OGIB. If the possibility of MD is already high with other modalities, patency CE may be an option before CE.

In the present case, it was difficult to diagnose MD before surgery. Several specific diagnostic tools have been reported to aid in the diagnosis of MD, including arteriography, technetium-99m pertechnetate scan, CE, and BAE. Among these, arteriography and technetium-99m pertechnetate scans are regarded as the standard tools for diagnosing MD. If bleeding is heavy, arteriography may be a good option. In patients with less bleeding, high-resolution CT arteriography can be a good option, and its overall sensitivity for detecting active acute GI bleeding is reported to be as high as 85.2\%.\textsuperscript{14} If bleeding stops spontaneously, it can be difficult to detect the source of bleeding using high-resolution CT arteriography. The technetium-99m pertechnetate scan failed to diagnose the presence of MD in the present case, although the heterotopic gastric mucosa was confirmed pathologically after surgery. The sensitivity of the technetium-99m pertechnetate scan for adult MD has been reported to be approximately 60%.\textsuperscript{7} Krstic et al. reported that CE had a high positive predictive value of 84.6\% for the diagnosis of MD.\textsuperscript{5} However, in the same study, CE identified the source of bleeding in only 44.6\% of patients with OGIB. Although several CE findings for the diagnosis of Meckel’s diverticulum have been reported, it is often difficult to diagnose Meckel’s diverticulum under physiological conditions without air inflation. Furthermore, control of CE movement is impossible, and it may pass through Meckel’s diverticulum. In the present case, several diagnostic tools, including high-resolution CT arteriography, technetium-99m pertechnetate scan, and CE, failed to diagnose MD. Considering the insufficient sensitivity of some tests to diagnose MD, several examinations, including BAE, should be performed to diagnose MD. Although BAE is an invasive procedure, it may be preferable to other modalities, especially for the diagnosis of MD.\textsuperscript{6,15}

In addition to the difficulty of MD diagnosis, we should keep in mind the possibility of intestinal stricture due to MD in patients with OGIB.
AUTHOR CONTRIBUTIONS
NS wrote the manuscript. KI, TT, SY, TG, FY, KK, MT, YK, AK, FY, YT, and HY reviewed and edited the manuscript. GT made the pathological diagnosis and reviewed the manuscript. MK, KY, and MM performed the surgery and provided detailed information about the procedure.

ACKNOWLEDGMENTS
We would like to thank Editage (www.editage.com) for English-language editing.

CONFLICT OF INTEREST
None declared.

ETHICAL APPROVAL
Written informed consent was obtained from the patients. Formal approval is not necessary for a case report in our hospital.

DATA AVAILABILITY STATEMENT
Data sharing is not applicable to this article as no new data were created or analyzed in this study.

CONSENT
Written informed consent was obtained from the patient to publish this report in accordance with the journal’s patient consent policy.

ORCID
Shinya Nakatani https://orcid.org/0000-0003-3498-5883

REFERENCES
1. Raju GS, Gerson L, Das A, Lewis B. American Gastroenterological Association. American Gastroenterological Association (AGA) institute medical position statement on obscure gastrointestinal bleeding. Gastroenterology. 2007;133:1694-1696.
2. Liao Z, Gao R, Xu C, Li ZS. Indications and detection, completion, and retention rates of small-bowel capsule endoscopy: a systematic review. Gastrointest Endosc. 2010;71:280-286.
3. Shinozaki S, Yamamoto H, Yano T, et al. Long-term outcome of patients with obscure gastrointestinal bleeding investigated by double-balloon endoscopy. Clin Gastroenterol Hepatol. 2010;8:151-158.
4. Hansen CC, Soreide K. Systematic review of epidemiology, presentation, and management of Meckel’s diverticulum in the 21st century. Medicine. 2018;97:e12154. doi:10.1097/MD.0000000000012154
5. Krstic SN, Martinov JB, Sokić-Milutinović AD, Milosavljević TN, Krstić MN. Capsule endoscopy is useful diagnostic tool for diagnosing Meckel’s diverticulum. Eur J Gastroenterol Hepatol. 2016;28:702-707.
6. Fukushima M, Kawanami C, Inoue S, Okada A, Imai Y, Inokuma T. A case series of Meckel’s diverticulum: usefulness of double-balloon enteroscopy for diagnosis. BMC Gastroenterol. 2014;14:155.
7. Lin S, Suhocki PV, Ludwig KA, et al. Gastrointestinal bleeding in adult patients with Meckel’s diverticulum: the role of technetium 99m pertechnetate scan. South Med J. 2002;95:1338-1341.
8. Routh WD, Lawdahl RB, Lund E, Garcia JH, Keller FS. Meckel’s diverticula: angiographic diagnosis in patients with non-acute hemorrhage and negative scintigraphy. Pediatr Radiol. 1990;20:152-156.
9. Adler DG, Knipschield M, Gostout C. A prospective comparison of capsule endoscopy and push enteroscopy in patients with GI bleeding of obscure origin. Gastrointest Endosc. 2004;59:492-498.
10. Rezapour M, Amadi C, Gerson LB. Retention associated with video capsule endoscopy: systematic review and meta-analysis. Gastrointest Endosc. 2017;85:1157-68.e1152.
11. Courcoutsakis N, Pitiakoudis M, Mimidis K, Vradelis S, Astrinakis E, Prassopoulos P. Capsule retention in a giant Meckel’s diverticulum containing multiple enteroliths. Endoscopy. 2011;43 Suppl 2:E308-E309.
12. Ben Soussan E, Antonietti M, Hervé S, et al. Diagnostic yield and therapeutic implications of capsule endoscopy in obscure gastrointestinal bleeding. Gastroenterol Clin Biol. 2004;28:1068-1073.
13. Sears DM, Avots-Avotins A, Culp K, Gavin MW. Frequency and clinical outcome of capsule retention during capsule endoscopy for GI bleeding of obscure origin. Gastrointest Endosc. 2004;60:822-827.
14. García-Blázquez V, Vicente-Bártulos A, Olavarria-Delgado A, et al. Accuracy of CT angiography in the diagnosis of acute gastrointestinal bleeding: systematic review and meta-analysis. Eur Radiol. 2013;23:1181-1190.
15. Hong SN, Jang HJ, Ye BD, et al. Diagnosis of bleeding Meckel’s diverticulum in adults. PLoS One. 2016;11:e0162615.

How to cite this article: Nakatani S, Inoki K, Tashiro T, et al. Small intestinal bleeding and stricture caused by Meckel’s diverticulum. Clin Case Rep. 2022;10:e06144. doi: 10.1002/ccr3.6144