



Case Report

Success of Minimally Invasive Transumbilical Single-Incision Laparoscopic Surgery (T-SILS) Plus Double-Balloon Endoscopy (DBE) for Pediatric Intestinal Angiodysplasia: A Case Report

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We describe here a child with angiodysplasia of the small intestine, diagnosed by double-balloon endoscopy (DBE), who was treated with transumbilical single-incision laparoscopic surgery (T-SILS). A 9-year-old boy presented to another hospital with intermittent fresh melena of a duration of 5 months and 4 days. Anoscopy and gastric mucosal membrane scintigraphy were unsuccessful diagnostically, and he was referred to our hospital for further examination and treatment. Under general anesthesia, we performed DBE by an anal route, with the ileum assessed for a distance of about 150 cm from the ileocecal valve. Although no bleeding lesion was found in the colon, a flat elevated venous lake approximately 20 mm in size was observed 20 cm from the ileocecal valve, suggesting angiodysplasia of the small intestine. A tattoo was made under DBE, and wedge resection by T-SILS was performed 1 month later under general anesthesia. The excised specimen appeared as a flat, elevated venous lake approximately 20 mm in size. Histopathologic analysis revealed several dilated and distorted veins within the submucosa of the small intestine. The patient was diagnosed with angiodysplasia of the small intestine. His postoperative course was uneventful, and he remained free of recurrent gastrointestinal bleeding 18 months after the operation. This is the first case report showing the diagnosis, localization, and tattooing of an obscure gastrointestinal bleeding lesion of the small intestine by preoperative DBE and removal of the lesion by subsequent T-SILS in children.

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Approximately 1% to 5% of patients with gastrointestinal bleeding can be classified as having obscure gastrointestinal bleeding (OGIB), defined as bleeding of unknown origin that persists or recurs after a negative initial or primary colonoscopy and/or upper endoscopy.^{1–3} Small intestinal angiodysplasias are found in 30% to 40% of patients with OGIB.^{4,5} Small intestinal bleeding can be detected, diagnosed, and localized by double-balloon endoscopy (DBE).⁶ No consensus has been reached regarding the most effective therapeutic strategy for angiodysplasia of the small intestine. We describe here a pediatric patient with angiodysplasia of the small intestine who was successfully treated by DBE combined with transumbilical single-incision laparoscopic surgery (T-SILS).

Case Report

A 9-year-old boy presented to another hospital with intermittent fresh melena 5 months and 4 days in duration. Anoscopy and gastric mucosal membrane scintigraphy were unsuccessful in diagnosing his condition, and he was transferred to our hospital for further examination. The patient had symptomatic epilepsy, cerebral palsy, and mental retardation, but no obvious abnormal physical findings on admission, including blood biochemistry. His symptoms suggested lower gastrointestinal tract bleeding, perhaps small intestinal bleeding. DBE (Fujinon EN-450p5/20, Fujinon Inc, Saitama, Japan) was performed under general anesthesia. Although no bleeding lesion was found in either the total colon or the ileum (150 cm from the ileocecal valve), a flat, elevated venous lake approximately 20 mm in size was found 20 cm from the ileocecal valve (Fig. 1A). We suspected angiodysplasia of the small intestine. The area around the lesion was tattooed endoscopically for subsequent laparoscopic surgery. Because of the possibility of repeated future gastrointestinal bleeding, surgery was performed 1 month later. The patient was placed under general anesthesia, and T-SILS was performed with the patient in a supine position. A reverse Ω -shaped incision was made at the umbilicus, and a multichannel EZ access (Covidien, Tokyo, Japan) was inserted as a platform under direct vision. A 5-mm flexible scope (Olympus, Tokyo, Japan) and 2 noncrushing graspers were

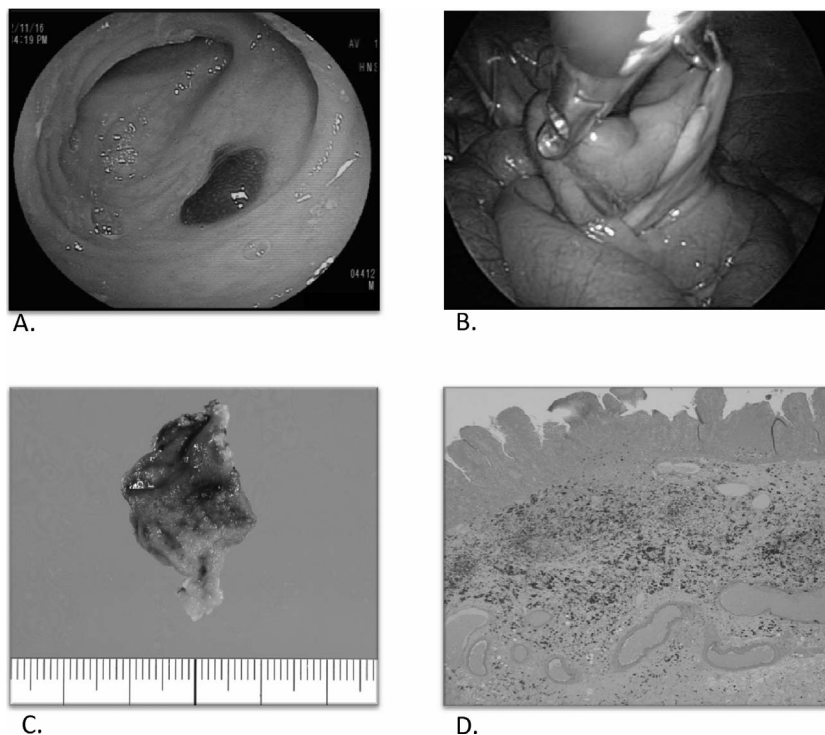
introduced into the EZ access. The previously tattooed small intestine lesion was immediately identified (Fig. 1B) and extracted from the abdominal cavity through the umbilical incision. A wedge resection was made around the tattooed site. The excised specimen appeared as a flat, elevated venous lake approximately 20 mm in size (Fig. 1C). Histopathologic analysis showed several dilated and distorted veins within the submucosa of the small intestine (Fig. 1D), resulting in a diagnosis of angiodysplasia of the small intestine. The patient's postoperative course was uneventful, and he was discharged 7 days after the operation. The patient remains free of recurrence 18 months after the operation.

Discussion

Angiodysplasia of the small intestine may be treated endoscopically, pharmacologically, surgically, or by interventional radiology. No consensus has yet been reached about the most effective therapeutic strategy for bleeding angiodysplasia of the small intestine. Although approximately 90% of hemorrhagic angiodysplasias spontaneously cease bleeding,⁷ the risk of recurrent bleeding is high. For example, the cumulative incidences of overt rebleeding at 1 and 3 years were 26% and 45%, respectively, and the rates of occult rebleeding at 1 and 3 years were 46% and 64%, respectively.^{8,9} A previous episode of rebleeding is an independent risk factor for further rebleeding.¹⁰ Up to 12% of these patients require surgical treatment,¹¹ and the angiodysplasia related mortality rate is around 2%.¹² Curative surgical resection is recommended for patients with gastrointestinal angiodysplasia. Lesions too small and too soft to be palpated may be difficult to localize intraoperatively, even during laparotomy. Thus, marking a lesion before surgery is important.

The clinical introduction of DBE markedly changed the diagnosis and treatment of small intestinal bleeding.¹³ DBE allows endoscopic imaging of the entire small intestine, an organ inaccessible using conventional procedures, allowing the bleeding site to be diagnosed and localized. DBE was able to diagnose 64% of patients with small intestinal OGIB who could not be given a diagnosis

Fig. 1 Histologic appearance of the lesion in our patient. (A) A flat, elevated venous lake approximately 20 mm in size and located 10 cm from the ileocecal valve. (B) Laparoscopic view of the tattoo of the lesion in the ileum, enabling easy identification of the lesion during subsequent laparoscopic surgery. (C) Photograph of the excised specimen, showing lifting of a 20-mm area of the mucous membrane. (D) Several dilated and distorted veins present within the submucosa of the small intestine. Hematoxylin-eosin ($\times 40$).



by conventional approaches.¹⁴ We found that DBE could successfully give a diagnosis of OGIB and localize its site of origin in our patient. Labeling the lesion allowed its later surgical removal.

Surgical methods for small intestinal diseases have changed since the introduction of DBE. As in our patient, an integrated approach consisting of DBE and laparoscopic bowel surgery (LBS) was used to manage small intestinal diseases manifesting as OGIB and vague abdominal pain when nonoperative methods failed or were not appropriate. LBS plus DBE has advantages over laparotomy without DBE. First, the surgical incision is smaller in LBS than in laparotomy, reducing postoperative pain. Moreover, LBS is faster, safer, has better cosmetic outcomes, and is more cost-effective than laparotomy. Most laparoscopically assisted intestinal resections can be completed in 1 hour, with no mortality and minimal morbidity.¹⁵ Second, the use of LBS in a targeted fashion diminished the need for thorough exploration and manipulation of the entire intestine. In most cases, intestinal targets were easily identified at initial laparoscopic examination based on the “tattoo” procedure. Third, by exfoliating the adhesion laparoscopically, it is easy to displace the organ outside the abdominal cavity to remove any physiologic adhesion, such as in the ascending colon

or the ligament of Treitz. Exteriorization of the diseased intestinal loop at the nearest port site allowed any surgical procedure to be easily performed.

Resumption of bowel movement, surgical morbidity and mortality rates, and hospital stay of patients managed using LBS plus DBE compared favorably with those of patients treated with exploratory laparotomy and intraoperative endoscopy.^{2,16,17} T-SILS was minimally invasive, allowing good outcomes in our patient.

To date, 26 children with small intestinal disease have been managed by SILS.^{18–23} Many diseases are not accompanied by a specific lesion site, such as Meckel diverticulum and Crohn disease, making preoperative identification of any lesion important. To our knowledge, this is the first case report showing the diagnosis, localization, and tattooing of an OGIB lesion of the small intestine by preoperative DBE, and removal of the lesion by subsequent T-SILS.

In conclusion, DBE is useful in the diagnosis and localization of lesions in the small intestine. DBE plus T-SILS enabled the successful management of this OGIB in a child using minimally invasive laparoscopic surgery. The combination of DBE plus

T-SILS may be a standard approach to OGIB lesions in the small intestine.

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