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Hemorrhagic Meckel's diverticulum in an older woman diagnosed by repeated angiographies

Erişkin kadın hastada anjiografik incelemeler ile tanı konan hemorajik meckel divertikülü

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Meckel's diverticulum is a remnant of the vitelline duct and a congenital anomaly of the gastrointestinal tract. Hemorrhage from a Meckel's diverticulum is common in children but extremely rare in adults over 50 years of age. Very few cases have been reported to date and all prior cases were in men. Meckel's diverticulum is commonly overlooked as a possible cause of a lower gastrointestinal hemorrhage in adults. Here, we present the rare case of a 58-year-old woman with massive hemorrhage from a Meckel's diverticulum that was diagnosed by repeated emergency angiographies and treated with elective laparoscopic surgery.

Key words: Meckel diverticulum, hemorrhage, angiography, laparoscopy

Meckel divertikülü, vitellin kanalın artığıdır ve gastrointestinal sistemin konjenital anomalileri arasında yer almaktadır. Meckel divertikülünden kanama çocuklarda sık karşılaşılan bir durum olmasına karşılık, bu durum 50 yaşını aşmış erişkinlerde nadirdir. Günümüze kadar oldukça az sayıda vaka bildirilmiştir ve bundan önce yayınlanmış vakaların hepsi erkektir. Burada, massif gastrointestinal kanama nedeni ile acil koşullarda yapılan birçok anjiografik inceleme sonucunda hemorajik Meckel divertikülü tanısı alan 58 yaşındaki kadın hasta sunulmuştur.

Anahtar kelimeler: Meckel divertikülü, kanama (hemoraji), anjiografi, laparaskopi

INTRODUCTION

Meckel's diverticulum is a remnant of the vitelline duct and the most common congenital anomaly of the gastrointestinal tract (1). It occurs in 2-3% of the population and rarely becomes symptomatic; it has a lifetime complication rate of 4-6% (2, 3). Among patients with symptomatic Meckel's diverticulum, the male-female ratio has been reported to be approximately 3:1 (3). The major complications include inflammation, hemorrhage, intussusception, small bowel obstruction, and neoplasm (2, 3). Hemorrhage from Meckel's diverticulum is common in children but extremely rare in adults over 50 years of age (4-6).

Meckel's diverticulum is usually overlooked as a possible cause of lower gastrointestinal hemorrhage in adults (4-6). We report the case of a 58-year-old woman with a gastrointestinal hemorrhage secondary to a Meckel's diverticulum diagnosed by repeated emergency angiographies and treated with elective laparoscopic surgery.

CASE REPORT

A 58-year-old woman was admitted to our department on an emergency basis because of a sudden onset of hematochezia immediately after straining with a normal bowel movement. The patient denied abdominal pain and prior episodes of bloody stools. There was no significant medical or family history. There was no history of alcohol intake, or non-steroidal antiinflammatory drug (NSAIDs) or tobacco use. The patient had a resting tachycardia, but blood pressure was normal. No abnormal findings were noted on the physical examination or laboratory tests except for slightly increased bowel sounds and a decreased hemoglobin (9.3 g/dl). Nasogastric lavage yielded clear gastric fluid. Two

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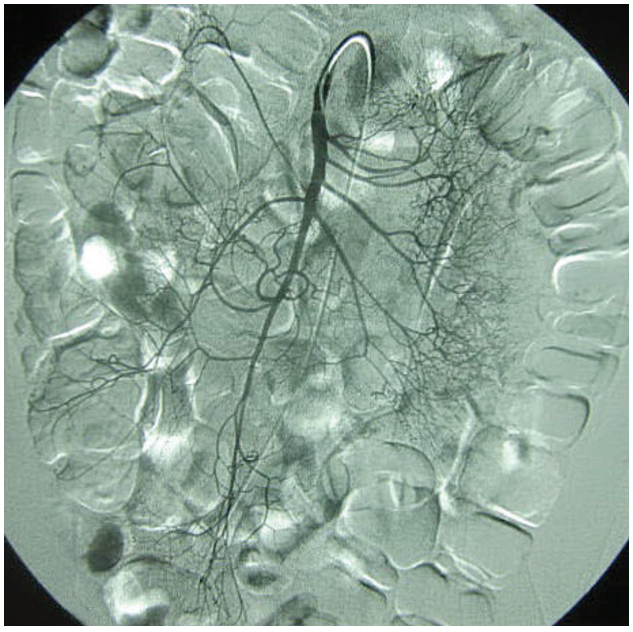

Figure 1. Initial angiography of the superior mesenteric artery shows no definite evidence of abnormal vasculature or extravasation focus.

units of packed red blood cells were immediately transfused and an urgent colonoscopy was performed without bowel preparation. A large amount of dark blood and clots were present in the entire left colon and rectum; the right colon could not be examined due to the presence of excessive blood.

The following day, the patient had another episode of massive hematochezia and the hemoglobin dropped even further to 5.6 g/dl; her blood pressure dropped to 90/60 mmHg and a tachycardia of 110/min was present. The patient was transfused with six units of packed red blood cells and three units of fresh frozen plasma. A second colonoscopy without bowel preparation was performed. A large amount of fresh blood was present along the entire colon and flowed out from the terminal ileum; however, no definite bleeding focus could be found. Under 100/70 mmHg blood pressure, an emergency angiography of the superior mesenteric artery (SMA) was performed concurrently with the blood transfusion, but showed no abnormality (Figure 1).

A third episode of massive hematochezia occurred on the third hospital day. The hemoglobin was 7.8 g/dl. The blood pressure dropped to 85/55 mmHg and a tachycardia of 115/min was present. An immediate transfusion was required; however, there was no blood available specific for this patient in the blood banks within about 200 km of our hospital. Before taking the decision of the last exploratory laparotomy, as a final attempt at a diagnosis and treatment for the bleeding, a second emergency selective superior mesenteric angiography was performed. At first, the angiographic findings were negative. However, repeated infusions of in-

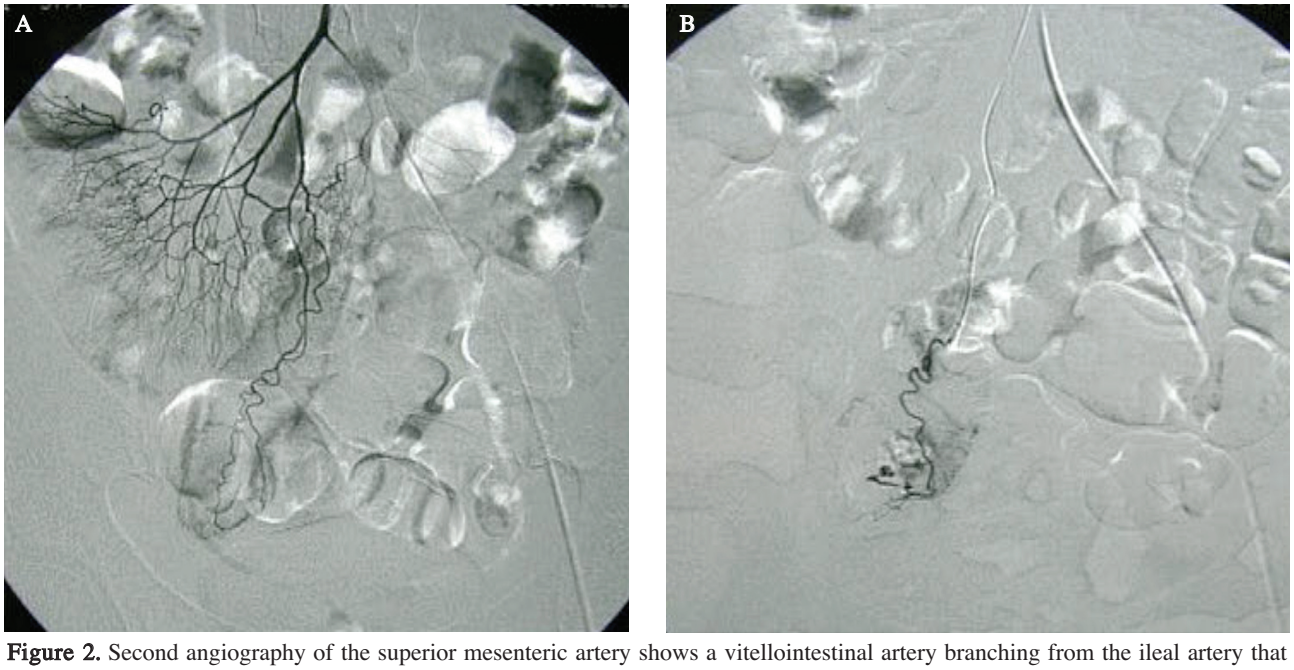

Figure 2. Second angiography of the superior mesenteric artery shows a vitellointestinal artery branching from the ileal artery that did not join with other ileal branches **(A)** and extravasation from it **(B)**.

tra-arterial contrast medium concurrently with rapid intravenous infusion of normal saline (until reaching a blood pressure of 130/80 mmHg) revealed a single elongated, non-branching arcade of an abnormal ileal artery arising from the distal SMA with intermittent extravasations of the contrast medium (Figures 2A, 2B). This artery was thought to be the vitellointestinal artery, a feeding artery of Meckel's diverticulum. Immediate embolization was carried out successfully with Gelfoam particles and two coils. After the procedure, the vital signs promptly stabilized to 140/90 mmHg and the pulse rate to 76/min. For protection from recurrent bleeding, a curative elective laparoscopic surgery without blood transfusion was performed three hours later. A 4.0 cm-long and 2.0 cm-wide Meckel's diverticulum was noted at about 50 cm proximal to the ileocecal junction on the antimesenteric side; it was resected with a laparoscopic stapler (Figure 3A). Multiple hemorrhagic erosions and small ulcers were noted in the resected specimen (Figure 3B). The pathology findings confirmed the diagnosis of Meckel's diverticulum and heterotopic gastric mucosa with ulcers (Figure 4A, 4B). The patient had no postoperative complications and was discharged on postoperative day 7; there have been no further episodes of gastrointestinal bleeding over the one-year follow-up.

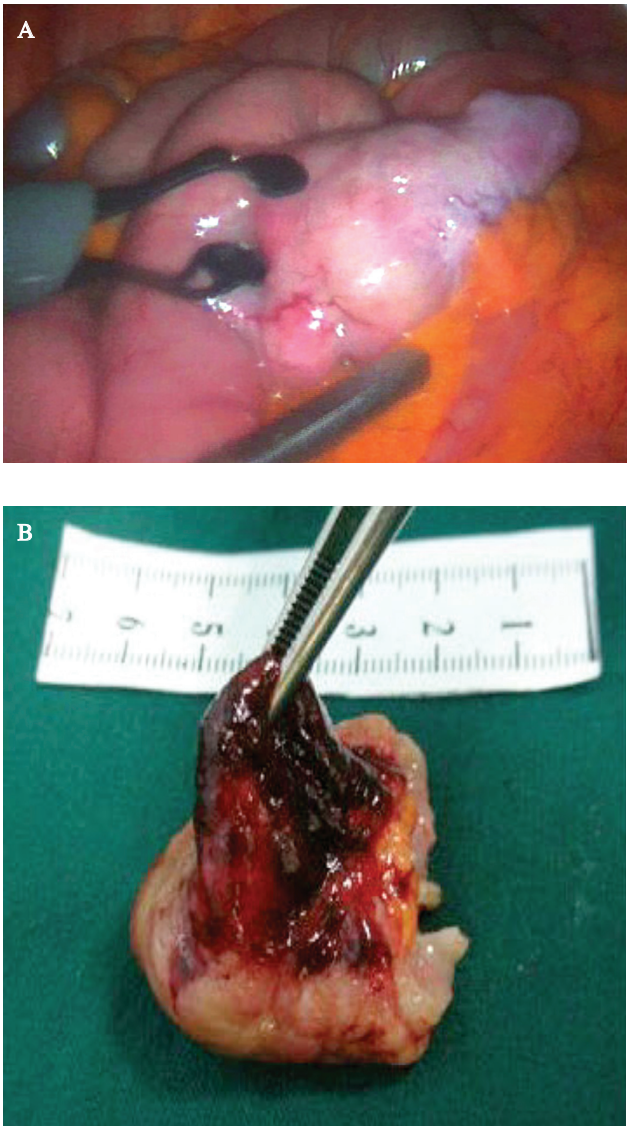

Figure 3. The findings of laparoscopic surgery show a 4.0 x 2.0 cm Meckel's diverticulum on the antimesenteric side of the ileum, 50 cm proximal to the ileocecal junction **(A)**. The opened and resected surgical specimen shows the hemorrhages in Meckel's diverticulum **(B)**.

DISCUSSION

The causes of obscure small intestinal hemorrhage in adults include angiodysplasia, a small bowel tumor, Crohn's disease, and NSAID-induced ulcers (7). However, hemorrhage from a Meckel's diverticulum is very rare in the adult age group (7). Mackey et al. (8) reviewed 402 patients with Meckel's diverticulum. Only 16.9% of the patients were symptomatic and one- quarter of them had gastrointestinal hemorrhage; the oldest was 31 years of age (8). There are only three cases of hemorrhagic Meckel's diverticulum that have been reported in patients more than 50 years of age, at 53, 68 and 91 years (9-11). They were all men. Therefore, this case is the first report of hemorrhagic Meckel's diverticulum in a woman over 50 years of age.

In our case, the diverticular wall had ectopic gastric mucosa with ulcerations. More than 80% of hemorrhagic diverticula contain ectopic gastric mucosa (4). This acidic environment may lead to ulcer formation within the diverticulum and this is the likely reason for gastrointestinal hemorrhage (4). The hemorrhage is usually painless, as in our case12. The hemorrhage can be massive and dramatic, manifesting as bright red blood in the stool, or it can be slow and occult, manifesting as anemia (12).

We performed repeated emergency angiographies under the unstable vital signs but did not consider Meckel's diverticulum as a possible cause of the massive hemorrhage since it would be extremely rare given our patient's age. Tc^{99m}pertechnetate scanning is known as one of the most useful diagnostic methods for the diagnosis of Meckel's diver-

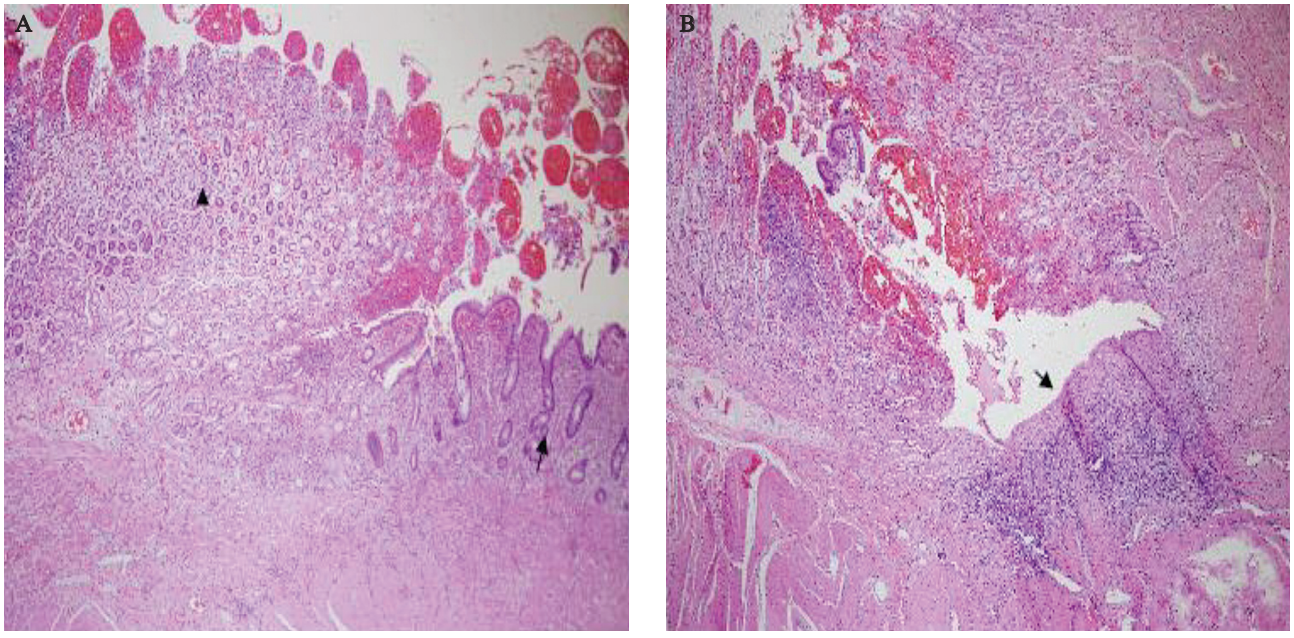

Figure 4. Microscopic findings of the resected specimen show hemorrhagic erosions in the mucosa with underlying ectopic gastric glands (arrow head) and adjacent to typical gastric epithelium, small intestinal epithelium (arrow) (hematoxylin and eosin [H&E], x40) **(A)**. Marked ulceration of the mucosal surface (arrow) with hemorrhage and inflammatory infiltration was noted (H&E, x40) **(B)**.

ticulum with heterotopic gastric mucosa (13). The diagnostic accuracy is more than 90% in children and only 50% to 65% in adults (13). However, this procedure cannot be performed in an emergency status, as was the case with our patient.

Angiography of the mesenteric artery is helpful during active gastrointestinal hemorrhage (14). The confirmation of the diagnosis of hemorrhage is made when contrast medium extravasation occurs into the bowel lumen. However, it is difficult to find the bleeding site when the rate of bleeding is less than 0.5 ml/min (14). However, in cases of Meckel's diverticulum, where the bleeding might be intermittent, the demonstration of an abnormal embryonic artery might be the most noticeable finding (15). Such a finding indicates the high probability of Meckel's diverticulum, even without extravasation, of a bleeding focus (15). The embryonic artery is characteristically a single, enlarged, elongated, unbranched anastomotic arcade originating from an ileal branch, or from the ileocolic artery of the distal SMA (16). In this case, even the abnormal vasculature was not visualized in the initial angiography under the low blood pressure. This suggests the probability of not only a lower rate of bleeding than 0.5 ml/min or intermittent bleeding (14), but also spasm of the abnormal feeding artery, which had a physiologic response to bleeding and low systemic intravascular volume with low blood pressure (15). We used the rapid intravenous infusion of normal saline to fill the intravascular volume and raise the blood pressure against the circulatory collapse. The diagnostic rate of angiography, for gastrointestinal hemorrhage, has been reported to be between 40% and 78% (17). Repeated angiography might be useful to localize a bleeding focus in patients with gastrointestinal bleeding of obscure origin who have a negative initial angiography (18). We successfully detected an abnormal feeding artery of Meckel's diverticulum by repeated angiography with rapid intravascular volume-filling in an extreme situation when no blood for transfusion was available.

In conclusion, we report the first case of a massive hemorrhagic Meckel's diverticulum in an older woman over 50 years of age, diagnosed by repeated emergency angiographies and treated with an elective laparoscopic surgery.

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