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Successful endovascular treatment of a 13-month-old child with gastrointestinal bleeding due to Dieulafoy syndrome of duodenum

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ABSTRACT

Dieulafoy disease can manifest itself with spontaneous massive recurrent gastrointestinal bleeding in children. We report a case of successful management of a 13-month-old child with Dieulafoy disease of duodenum when traditional methods of examination and treatment failed.

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In 1897, a French pathologist named Paul Georges Dieulafoy mentioned the term “exulceratio simplex” describing an atypical ulcer with a gaping arteriole in the submucosal layer of the stomach found at autopsy in 10 patients with an unexplained massive bleeding. Subsequently, a similar finding was reported in all regions of the gastrointestinal tract and in the bronchi [1]. The Dieulafoy disease manifests itself with spon-

aneous recurrent gastrointestinal bleeding (GIB). There is no objective statistics on the incidence of this disease in children to date. The Dieulafoy disease is observed in 6.7% of the cases of GIB in adults [2]. Available medical literature describes 28 cases of this disease in children [3]. Endoscopic, endovascular, and surgical treatment can be applied to adults; however, Dieulafoy disease located in the intestine is usually

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treated surgically [1]. Modern methods of interventional radiology, such as endovascular embolization, are increasingly employed in pediatric surgery. However, only 1 case of successful endovascular hemostasis of Dieulafoy disease, with localization in the duodenum, has been described in a 14-year old-child [4-7]. No employment of this method in children of younger age has been found. This article highlights a poorly diagnosed case of Dieulafoy disease located in the duodenum with a successful treatment in a 13-month-old child.

A boy aged 13 months with seizure, general weakness, and pale skin was admitted to the clinic of Saint Petersburg State Pediatric Medical University on February 26, 2013. According to his mother, the onset was sudden at 1 hour before their arrival at the clinic. No similar condition had been seen previously.

In the emergency department, the patient was examined by a pediatric surgeon (no acute surgical pathology was revealed), a pediatrician, and a neurologist who suspected a paroxysmal condition of unknown origin. The boy was admitted in the neurological department, where he underwent a neurologic examination (no focal or cerebral symptoms were found and the electroencephalogram presented no epileptic activity). The ultrasound scan of abdominal cavity showed moderate splenomegaly and reactive changes of the pancreas. There was nothing special on his electrocardiogram. His complete blood test results contained 10×10^9 109/L leukocytes, 2.8×10^{12} 1012/L erythrocytes, and 68 g/L of hemoglobin. Fever was noted on the second day and the child was transferred to the infectious department. On February 28, 2013, the child's condition became worse (anxiety, abdominal pain, and melena appeared). The patient was re-examined by a pediatric surgeon and an intestinal invagination was suspected. The child was transferred to the intensive care unit of the surgical department for further treatment. Objective examination determined the following status: the child's condition was of moderate severity, the skin was clean, pale, his abdomen was symmetrical, not swollen, and moderately painful on palpation in the mesogastric region, no mass was determined in the abdominal cavity, and there was some doubt whether any symptoms of peritoneal irritation were present. Applying a flatus tube, we obtained a black stool (melena) from the patient. Abdominal x-ray was performed and no abdominal fluid levels or free gas were found. Because of the patient's medical history and clinical findings and diagnosis of GIB, Meckel's diverticulum was suspected. Diagnostic laparoscopy and revision of the abdominal cavity were performed. No intraoperative pathology of the abdominal organs was identified. Esophagogastroduodenoscopy (EGD) and fibrocolonoscopy (FCS) were performed in the operating room, and no clear source of the bleeding was identified at the time of inspection. The investigation was carried out with technical difficulties due to poor resolution of the portable equipment.

On March 1, 2013, the boy's condition was stable with no hemodynamic disturbances. No clinical signs of any ongoing massive upper GIB were observed. The child showed no signs of worry, his abdomen was symmetrical and not swollen, his palpation was painless, and there were no peritoneal symptoms in all parts. He received blood transfusion and homeostatic and symptomatic therapy.

On March 3, 2013, blood appeared again in the patient's stool. EGD and FCS were repeated. According to the EGD, the most

likely source of bleeding was an erosive ulcerative process in the proximal part of the duodenum. According to the FCS, a left-sided nodular colitis was noted, the colon was examined up to the splenic angle, and further examination was not possible due to the presence of some remnants of a previous bleeding.

On March 5, 2013, the patient was transferred from the intensive care unit to the Surgical Department.

On March 6, 2013, the child's condition deteriorated again and clinical and laboratory signs of GIB were noted. An urgent EGD was performed and the source of bleeding was found to be an eroded vessel in the proximal part of the postbulbar region of the duodenum. The endoscopic examination revealed some signs of a vascular malformation (Dieulafoy disease). Endoscopic hemostasis (an injection of 1 mL of 5% glucose solution into the site of bleeding, clipping, and coagulation by installed clips) was performed.

During the first day after this procedure, the child's condition remained relatively stable, a moderate hemorrhagic discharge was noted, and a self-dependent scanty stool (melena) was obtained.

On March 7, 2013, 3 episodes of vomiting with scarlet blood occurred. The child expressed anxiety and some symptoms of hemorrhagic anemia could be seen. Migration of endoscopic clips was identified through the abdominal x-ray.

Because of the severity of the condition, clinical and laboratory data of recurrent GIB, and ineffectiveness of conservative and endoscopic methods of hemostasis, the child was transferred to the interventional radiology department for an angiography of abdominal vessels with a possible further endovascular embolization of the bleeding source.

General anesthesia was performed and local anesthesia of 1 mL of 1% lidocaine solution was injected at the side of puncture of the right femoral artery, 21G needle was used for puncture. 4Fr introducer (Terumo Medical Corporation, Tokyo, Japan) was installed using Seldinger technique. The Pig Tail 4Fr catheter (Cordis Corporation, Miami Lakes, FL) was inserted into the abdominal aorta, aortography was performed in the anterior-posterior projection, and the patient's abdominal aorta and main vessels were not changed (Fig. 1). The catheter was replaced by a "Judkins Right coronary catheter 4Fr" (Cordis Corporation) and it was inserted into the gastroduodenal artery (GDA) of the patient. Selective digital subtraction angiography was performed where the dilated GDA was seen to anastomose with the superior mesenteric artery and the right gastro-omental artery. After selective angiography of the GDA was performed, the duodenal vasculature was visualized without any pathologic arteriovenous shunting. Some extravasated contrast agent was clearly noted in the duodenal lumen in the area of the bulb (Fig. 2). Furthermore, a Transit 2.8 Fr microcatheter (Codman Neuro, Raynham, MA) was advanced through the diagnostic catheter and was installed into the branch of the GDA supplying the duodenal bulb. Endovascular embolization was performed using 3 coils of Trufill with a diameter of 0.021 inch (Codman USA) installed as a single conglomerate to the duodenal branch of the GDA (Fig. 3). Control angiography of the common hepatic artery and superior mesenteric artery was then performed. The control angiography after embolization showed that GDA was occluded from the origin of the right gastro-omental artery, and there was a moderate collateral opacification of duodenal vasculature.

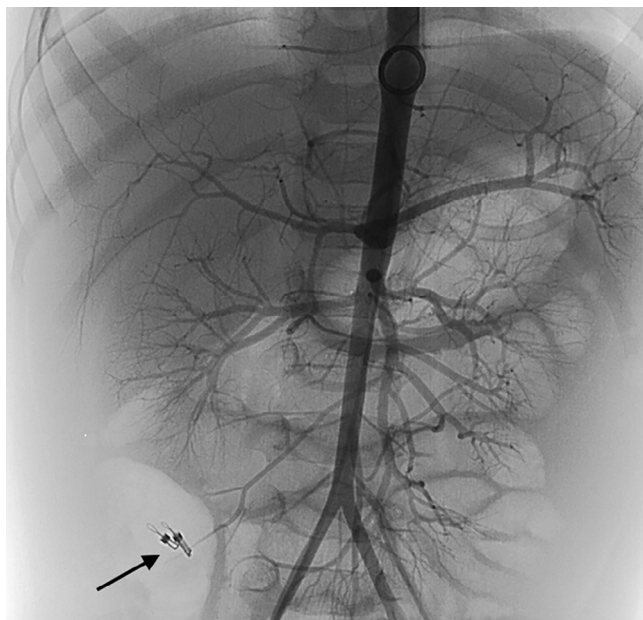


Fig. 1 – Abdominal aortography. No clear signs of extravasation. Migrated endoscopic clips shown with black arrow.

Embolization with particles was rejected due to the high risk of an ischemic lesion of duodenum. EGD was performed in the interventional radiology department and there were no data of an ongoing bleeding. Angiographic tools were removed and hemostasis was achieved by pressure to the site of puncture of the femoral artery. No complications were noted in the post-operative period. The condition of the child improved during the period of dynamic observation; enteral nutrition was extended and the child passed a daily independent stool without any pathologic impurities. There were no signs of recurrent GIB or any ischemia of the intestine. (Fig. 4)

On March 14, 2013, the boy was transferred to the surgical department, and on March 20, 2013, the child was discharged from the hospital.

The child was healthy with no signs of GIB identified during the 5-year follow-up.

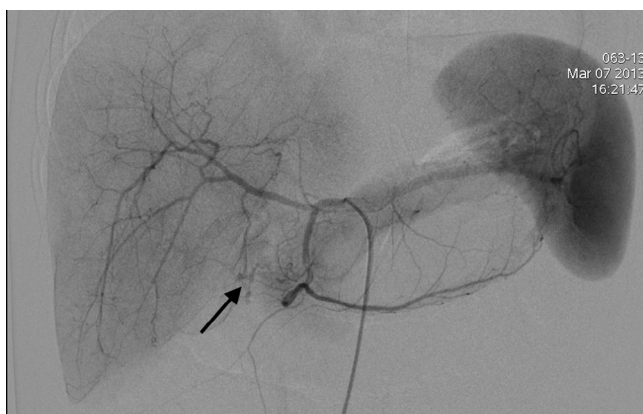


Fig. 2 – Selective celiac artery angiography—extravasation in duodenum shown with black arrow.

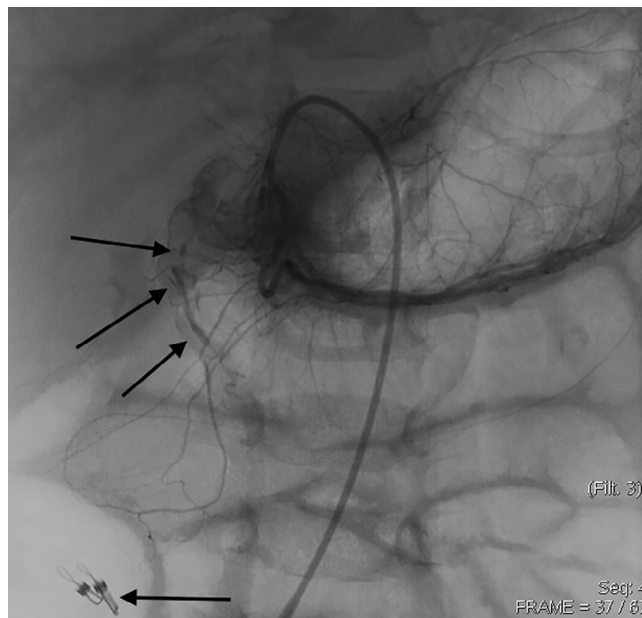


Fig. 3 – Selective angiography of gastroduodenal artery. Massive extravasation in duodenum shown with upper black arrows. Migrated endoscopic clips shown with lower black arrow.

Discussion

This case is interesting because in contrast with the performed traditional methods of examination (clinical and endoscopic ones and laparoscopy with revision of the abdominal cavity) and treatment (conservative therapy and endoscopic hemostasis), endovascular method proved to be most informative, highly efficient, and low-impact in the diagnosis and treatment of a child aged 13 months, with the Dieulafoy disease in the duodenum.

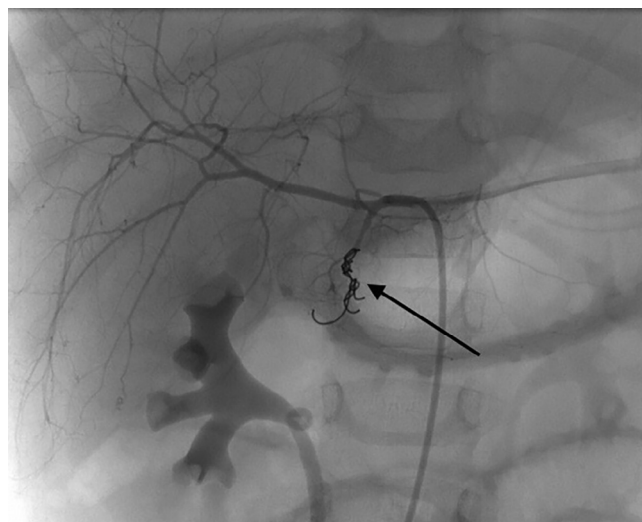


Fig. 4 – Celiac artery angiography after embolization. No signs of extravasation. Embolization coils shown with black arrow.

We have chosen microcoils as an occlusive material as their use does not lead to embolization of the distal vessels of the gut or to any ischemia. On the contrary, microcoils block the lumen of the artery and its branches of the first order, reducing the pressure of blood in the region of Dieulafoy ulcer, which is similar in effect to an extended surgical ligation of an artery. The method of endovascular embolization of gastrointestinal vessels with massive GIB can be applied in children of any age and can be an alternative to a surgical intervention.

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