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Successful diagnosis and treatment of Dieulafoy's lesion with endoscopy and thermocoagulation in a full-term neonate: Report of a case and literature review☆

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ABSTRACT

Dieulafoy's lesion is a rare cause of gastrointestinal tract hemorrhage in children. Herein, we report on a neonate presented with a massive upper gastrointestinal hemorrhage in the first postnatal day, successfully diagnosed and treated with endoscopy.

Upper gastrointestinal tract (UGIT) bleeding in the full-term newborn is not common. In a case–control study among 5180 healthy full-term newborns only 64 (1.2%) suffered from UGIT bleeding [1]. The most common causes included esophagitis, gastric ulcers, and gastric erosions. Dieulafoy's lesion (DL), first described extensively in 1898 [2], is characterized by the presence of a relatively large submucosal arterial vessel protruding through a solitary, small mucosal defect, located usually in the UGIT [3]. The lesion may rupture spontaneously causing a massive life-threatening hemorrhage [3]. Herein, we report on a neonate presented with hemorrhage of the UGIT, who was successfully diagnosed with endoscopy and treated with endoscopic thermocoagulation. A brief pediatric literature review is also cited.

1. Case report

This one-day male newborn was evaluated at the NICU 4 h after birth for massive bleeding from the UGIT. The boy was born at full term after Cesarean section due to cephalopelvic disproportion. His birth weight was 2.960 kg, and he had an Apgar score 9 at 1 and 10 at 5 min. No family history for bleeding disorders was reported. The baby was transferred to the maternity unit to start breast feeding where he vomited a small amount of bright-red blood. His vital signs were normal, with no delayed capillary refilling time. A nasogastric tube was placed and aspirated small amount of fresh blood with clots. Feeding was stopped, and ranitidine (2 mg/kg/day) was initiated. Initial laboratory tests were as follow: hemoglobin, 11.8 g/dL, (13.4–19.5 g/dL) hematocrit 38.5% (40–64%), leukocytes 20.400/μL (9000–30,000/μL) and platelets 359.000/μL (normal: 220–420.000/μL). Electrolytes and coagulation profiles were within normal range. A second big emesis consisted of bright-red blood was noticed 10 h later followed by passage of melanotic stools. The baby became pale with a heart rate of 170 beats per minute, blood pressure of 68/44 mm Hg, maintaining however appropriate capillary refilling time. Hematological measurements revealed a further drop of hemoglobin and hematocrit (9.3 g/dL and 30.1% respectively). After transfusion of one unit of packed red cells and one unit of fresh frozen plasma the infant was taken to the operating room for a UGIT endoscopy under general anesthesia. A neonatal endoscope Olympus® GIF-N180, external diameter 4.9 mm and operating channel of 2 mm, were used. After washing clots of blood away an oozing was visualized originating from a protruding small vessel measuring about 1 mm in diameter, and located at the lesser curvature of the stomach (Fig. 1). The nearby mucosa appeared normal.

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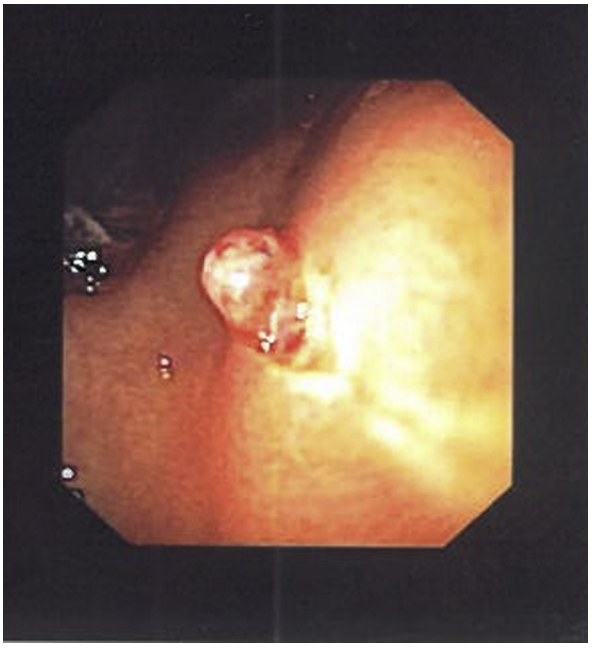

Fig. 1. Endoscopic view of the protruding vessel in the lesser curvature of the stomach in DL.

A metallic catheter was advanced through the operating channel and local tamponade was performed along with monopolar thermocoagulation. The bleeding stopped with no recurrence after a 10 min observation. A complete healing of the DL was noticed on a follow-up endoscopy after 18 days (Fig. 2).

2. Discussion

Adult literature estimates the prevalence of DL in UGIT bleeding between 0.3 and 6.7% [3]. In children it seems to be rare [4]. Using PUBMED and SCOPUS, we retrieved only 27 pediatric patients (aged 0–14 years), from the English literature (Table 1). Among them, there were only two neonates: the first involved a 3-day-old neonate [5] and the second a 4-day-old neonate [6]. Our case represents the earlier reported DL in children, since it was diagnosed within the first day of life, at approximately 16 h of life.

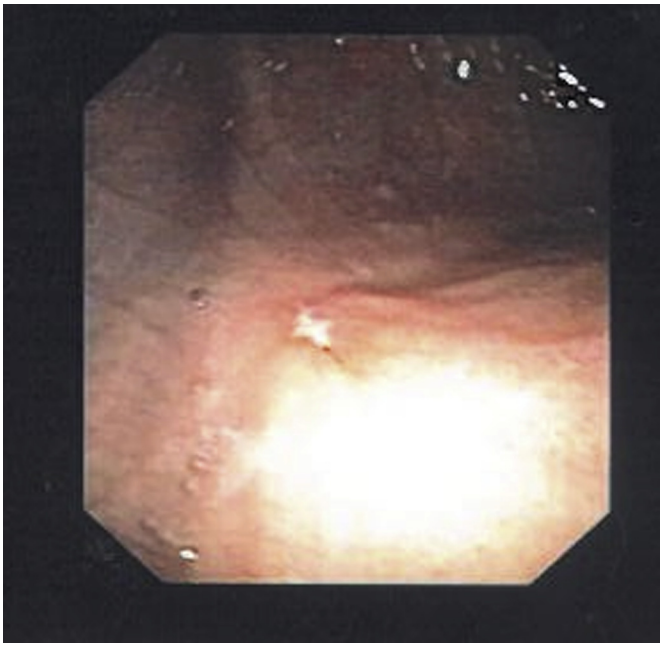

Fig. 2. Follow-up endoscopy 18 days after endoscopic thermocoagulation. No bleeding is seen.

Typically, a DL is located in the proximal portion of the stomach [5]. Occasionally, DL has been detected through-out the gastrointestinal tract [5] and the bronchi [4]. Of the listed pediatric cases, 15/27 (55.5%) were located in the stomach, two in the duodenum, five in the jejunum, two in the ileum and rectum respectively, and one in the sigmoid colon. Interestingly, in one patient, two simultaneous DLs, one in the stomach and another in the jejunum, were found (case 20).

The pathogenesis of DL is currently thought to be of congenital origin, as it has been found in neonates and infants [5–7]. Clinically, DL is presented as intermittent massive gastrointestinal bleeding, with no preceding symptoms. A predilection in men as compared to women has been noticed in adult series [4]. Probably, the presence of estrogen may play a protective role [4]. However, in children, a slight preponderance of young girls (1.2:1) was noted.

Since 1990s, advances in pediatric gastroenterology allow endoscopy for diagnostic procedures of the gastrointestinal tract even in neonates. In the listed cases, excluding our own, a diagnosis of DL was made endoscopically in 16/27 (59.25%) children. Among them, there were two neonates, and an 8-week-old infant. The following endoscopic criteria are acceptable for a definite diagnosis of the DL: a) active arterial spurting or micropulsatile streaming from a mucosal defect <3 mm or through normal surrounding mucosa, b) visualization of protruding vessel with or without bleeding, within a minute mucosal defect or through normal surrounding mucosa, and c) the appearance of fresh, densely adherent clot with a narrow point of attachment to a narrow mucosal defect or to normal appearing mucosa [8] However, repeated endoscopies may be required to identify the lesion because hemorrhage is often intermittent, and the site of bleeding may be covered by clots [5]. In the case that a DL is located at sites which are difficult to be approached by endoscope, other diagnostic modalities, such as selective angiography (cases 9, 20, 24, 27) (Table 1) and capsule endoscopy (case 18) (Table 1) may provide useful information.

Available data from the literature report a successful rate with endoscopic treatment $>90\%$ in adults with DL [9]. However, in the reviewed pediatric cases an endoscopic hemostasis was achieved only in 10 (37%) patients (Table 1). Endoscopic means included hemoclip, band ligation, sclerosants, epinephrine/norepinephrine injection, or combination. Stockwell [7] used sclerotherapy for the treatment of a 8-week infant with DL on the basis that thermocoagulation could cause perforation of the thin wall of the stomach. However, the definite method of treatment depends on the experience of the endoscopist and the availability of means of hemostasis. In the present case, we achieved hemostasis by using monopolar thermocoagulation. Since bleeding stopped, we elected not to infuse hemostatic agents afterward. Although surgery has now been reserved for the 5% of adults with DL [9], in the listed cases, the percentage of children treated surgically, either open or laparoscopically, was as high as 48% (Table 1). Reasons for that include studies reported before introduction of endoscopy in pediatric gastroenterology, and the localization of DL at sites that are not accessible with endoscopy such as the jejunum and ileum. However, the small number of pediatric cases does not allow the extraction of reliable conclusions. It is worth mentioning that a child with DL (case 27) was treated with embolization [10]. Similarly to adult series [3], recurrence of bleeding was low and noted only in one patient [(case 16) (3.8%)] successfully treated by a second endoscopy.