Duodenal and retroperitoneal hematoma after upper gastrointestinal endoscopy: First presentation of a child with Hemophilia B

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Abstract

Duodenal hematoma is an uncommon complication after upper gastrointestinal endoscopy. Most reported cases resolve with conservative management for 3–4 weeks. The authors report a case of a 7 year-old child who developed duodenal and retroperitoneal hematomas, as well as intra-peritoneal hemorrhage after a diagnostic upper gastrointestinal endoscopy. Surprisingly, this was the patient’s first presentation of his later-diagnosed Hemophilia B. This is the first Case report of duodenal hematoma after upper endoscopy as the only first presentation of a coagulation disorder. The case report also describes successful management of such complication.

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1. Case presentation

A 7-year-old male underwent upper and lower endoscopies for evaluation of failure to thrive and constipation. No family or personal history of bleeding disorder or easy bruising was declared by parents on pre-procedure assessment. Endoscopy showed no evidence of inflammation or mucosal abnormalities. Biopsies were taken from the proximal and distal esophagus, antrum, duodenum, terminal ileum, cecum, hepatic flexure, transverse colon, descending colon, and recto-sigmoid colon. During the procedure, there was no evidence of bleeding or mucosal friability, and there was good hemostasis after pinch biopsies. After the procedure, and while in recovery, the patient developed respiratory distress that required a nebulized bronchodilator treatment. Over-the-night stay in the hospital was complicated by several episodes of non-bloody non-bilious vomiting, decreased urine output, intolerance to oral intake, and abdominal pain in the suprapubic, and right lower quadrant regions. He developed hypotension, and altered mental status and was transferred to the pediatric intensive care unit (PICU), where he was electively intubated. His laboratory workup revealed metabolic acidosis, severe anemia, prolonged prothrombin time (PT), and prolonged International Normalized Ratio (INR). He was resuscitated with packed red blood cells (PRBCs), and fresh frozen plasma (FFP) transfusions. There was no sanguineous output through the nasogastric (NG) tube, and there was no blood reported in the stools. Plain X-ray film showed no free air in the peritoneal cavity. Limited ultrasound of the abdomen showed free fluid in the peritoneal cavity. Ultrasound-guided diagnostic paracentesis (DP) showed blood in the peritoneal cavity. The surgical team was consulted, and due to the degree of abdominal distension and the hemodynamic instability of the patient, exploratory laparotomy was favored over diagnostic laparoscopy as the mode of surgical intervention.

Exploratory laparotomy revealed a large duodenal hematoma extending from the second part of the duodenum to the ligament of Treitz, intra-peritoneal hemorrhage, and a retroperitoneal hematoma. The duodenum was dusky, and had an ischemic appearance, so, due to the concern that a serosal incision would cause a full...
thickening injury to the duodenum, evacuation of the hematoma was deferred. Jejunum was divided, and a proximal jejunostomy was brought to skin level, with a drainage tube passed retrograde toward the duodenum for drainage distal to the hematoma site. A feeding tube was placed into the distal jejunum, which was brought to skin level as a separate stoma, for enteral feeding. A gastrostomy tube was also placed for drainage proximal to the duodenal hematoma site. Two days later, the patient had a follow up exploratory laparotomy that showed improvement in the bowel color but no improvement in the duodenal hematoma size. Mucosal biopsies were all histologically normal. Early feeds were started through the distal jejunostomy and he was discharged home on J-tube feedings and re-feeding the gastric tube output into the jejunostomy. Seven days post-discharge he developed a hematoma at the site of the radial arterial line placement that required evacuation in a local emergency department (ED).

During the outpatient follow up visits, he was found to have persistently prolonged partial thromboplastin time (PTT), and mildly elevated PT and INR. The patient did not have any history of easy bleeding or bruising and the family denied any history of bleeding. Eventually, mother reported that she was anemic and was being evaluated for menometrorrhagia. Evaluation of his coagulopathy was delayed due to the multiple blood transfusions he had received. Once laboratory studies were obtained he had a normal Factor VIII level, normal von Willebrand factor (VWF) level, Factor XII level was 44% of normal, and Factor IX level was profoundly low at 2% of normal. These results were consistent with the diagnosis of moderate-severe Hemophilia type B, and he was subsequently started on Factor IX replacement therapy. The patient continued to gain weight through jejunostomy feeds, and 7 months post diversion he underwent successful jejunal re-anastomosis. He had an uneventful post-operative course and rapidly tolerated oral feeds.

2. Discussion

The incidence of occurrence of a duodenal hematoma after upper endoscopy is not well defined in the literature. It was shown in one study to be as high as 0.01% [1], while another showed the incidence to be as low as 1.1250 (0.0008%) [2]. In a prospective study among 1120 patients who had upper gastrointestinal endoscopy done, none developed duodenal hematomas [3].

The second and third portions of the duodenum are naturally prone to injury due to retroperitoneal fixation and limited mobility. Proposed mechanisms for the development of duodenal hematoma post endoscopy include: (a) Excessive manipulation of the tip of the endoscope inside the duodenum causing shearing and pressure forces that can damage the rich submucosal vascular plexus of the duodenum allowing the occurrence of a hematoma [4], (b) Tenting of the duodenal mucosa while obtaining biopsies. It has been recommended that the tip of the scope should not be more than 3 cm from the mucosa when obtaining biopsies to avoid excessive tenting of the mucosa by the biopsy forceps [5]. And (c) Impaired coagulation, or low platelet count. There are case reports of patients who have coagulation defects, or were on anticoagulant therapy developing duodenal hematoma either spontaneously [6,7], or after an upper endoscopy [8]. There are other case reports of patients with low platelet counts due to blood dyscrasias, or post-bone marrow transplant chemotherapy who also sustained an intramural hematoma of the duodenum after an upper endoscopy [9,10]. Sometimes, no risk factors can be identified, with the patients having normal coagulation profile, bleeding time, and normal platelet counts [11].

The clinical presentation of post-endoscopy duodenal hematoma includes: severe abdominal pain, bilious vomiting from duodenal obstruction, and intolerance to oral intake. Physical exam might show abdominal tenderness and distension [11]. Diagnosis of duodenal hematoma is predominantly by imaging. Several imaging modalities have been used; most notably ultrasound (US), computed tomography (CT) scan, Magnetic Resonance Imaging (MRI) and upper gastrointestinal (UGI) series. CT scan is usually the first choice for initial diagnosis as it delineates the extent of the hematoma, as well as the presence of free intraperitoneal air from bowel perforation. US is an alternative option for initial diagnosis, but it cannot accurately assess the extent of the hematoma or localize a perforation. It is most useful for following the resolution of the hematoma during conservative treatment [12]. MRI is another useful tool that is superior to CT scan due to less radiation exposure [11] but has longer scan time. Upper GI series may show a show “stacked coin” or “coil spring” appearance due to mucosal thickening [13]. No comparative study has been done to compare the advantage of one imaging study over the other [11]. Laboratory studies which are important in follow up include assessment of bilirubin and pancreatic lipase levels as the hematoma can obstruct the ampulla of Vater.

Management of post-endoscopy duodenal hematomas is mainly conservative and includes nasogastric (NG) aspiration, intravenous fluids, total parenteral nutrition (TPN) and Nothing per Os (NPO) status. Surgical intervention, once the most common intervention, is reserved to complicated cases as in the case of bowel perforation, or cases that did not resolve after conservative management [11,14]. Use of endoscopic hemoclips was recently reported to stop the bleeding from biopsy sites in the duodenum and stomach that caused intramural hematoma [15]. This might be challenging in younger infants due to the lack of availability of small sized clips. Evacuation of the hematoma can be done by open laparotomy, or laparoscopic assisted [15]. US [16] and CT [17] guided drainage has also been reported to be successful.

Our case had a significant surgical intervention. The duodenal hematoma was so massive that the surgeon determined that the resolution time would be quite prolonged, thus to avoid long term total parental nutrition, the decision was made to establish a feeding jejunostomy through which early tube feedings could be initiated in the early postoperative period.

The most common complication of duodenal hematoma is acute pancreatitis secondary to amputillary obstruction. It is noted to be more common in post endoscopy duodenal hematoma, than in post-traumatic hematoma, and usually improves with resolution of the duodenal hematoma [11]. Other complications include: jaundice due to obstruction of the biliary ducts, perforation, hemoperitoneum, and intestinal obstruction. Stricture formation is unusual with or without surgical intervention [18,19]. Death was reported in two leukemic patients with duodenal hematoma post-upper endoscopy, but, it was related to the primary illness, rather than due to complications of the hematoma [9,20].

3. Conclusion

Our case is an unusual presentation of Hemophilia B. There was no personal or obtainable family history of easy bleeding or bruising. Although this is an unfortunate case, due to the rarity of this condition and the cost of testing, we do not recommend routine laboratory screening prior to upper endoscopy. History and physical examination remain the most sensitive and cost-effective method for detecting bleeding disorders in pediatric (and adult) patients.
Conflict of interest statement
All authors declare that there are no conflicts of interest to disclose.

References