Unusual Presentation of Duodenal Dieulafoy's Lesion in a Toddler

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ABSTRACT

Dieulafoy's lesion is a rare cause of massive upper gastrointestinal haemorrhage in any age. It predominantly occurs in the proximal stomach. We report on a child who presented with massive rectal haemorrhage and a clear nasogastric aspirate due toduodenal Dieulafoy's lesion.

Keywords: *clear nasogastric aspirate; hematemesis; massive rectal haemorrhage; vascular malformation*

INTRODUCTION

Localisation and evaluation of gastrointestinal (GI) haemorrhage in clinical medicine is based on history and nasogastric aspirate. Bright red blood per rectum and a clear nasogastric aspirate suggest a lower gastrointestinal source and a massive bleed in a young child is commonly from Meckel'sdiverticulum with ectopic gastric mucosa.[1] We report on a child who presented with skin rash, massive rectal haemorrhage and a clear nasogastric aspirate due toduodenal Dieulafoy's lesion for the challenges in diagnosis and management.

CASE REPORT

A three year old male was brought to the emergency with history of low grade fever, rash and loose stools of four days duration and massive rectal bleed one episode prior admission. The rash was generalized, raised, erythematous, pruritic and painful. He had received prednisolone for the same. On examination child was irritable and pale with pulse rate of 180/min and blood pressure 86/50 mmHg. No skin rash was noted. Abdomen was soft and non tender with increased peristalsis. No mass was palpable. Systemic and digital rectal examinations were normal. Nasogastric aspirate was clear. Child was stabilized with packed cell transfusions after obtaining hemoglobin which was 3.6 gm/dL. Other investigations revealed neutrophilic leukocytosis, normal platelet count, prothrombin, partial thromboplastintime,liver and renal function tests. There was no proteinuria or hematuria on urine analysis. Ultrasound abdomen showed dilated rectosigmoid and fluid filled bowel loops. A Technetium (Tc) 99^{m} scan showed no tracer uptake suggestive of Meckel's diverticulum.

As there was no rectal bleed after admission for 72 hours, child was started on oral sips. Child developed abdominal pain, urticaria and recurrent hematochezia. As colonoscopy did not show any active lesion, laparotomy was planned. On exploration intraluminal blood was seen distal jejunum onwards; there was no Meckel's diverticulum or serosal hemorrhages to suggest vasculitis. Transillumination of entire intestine showed no abnormal vessel or bleeding site. In the immediate post operative period, child developed hemetemesis and hematochezia. Bleeding was torrential and he was resuscitated with fluids, whole blood and vasopressin

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infusion. Upper GI endoscopy showed blood clots in stomach and duodenal bulb; blood was seen filling the stomach from below suggestive of small bowel bleed. On re exploration, stomach and entire small bowel were filled with blood; jejunotomy and intraoperative endoscopy could not identify the bleeder. Duodenal filling was noted following Kocherisation. Following duodenotomy a solitary pin point arterial spurter (Dieulafoy's lesion) was identified on the medial surface of the first part of the duodenum with normal surrounding mucosa. The vessel was under run both on the mucosal and serosal surfaces. Child was electively ventilated for 36 hours post operatively. During endoscopy and re-exploration, volume replacement included blood products and intravenous fluid amounting approximately to four times his estimated blood volume. Coagulopathy in the post operative period was managed with fresh frozen plasma. Feeds were started on 7th post operative day and subsequently he had an uneventful recovery.

DISCUSSION

First described in 1897 by a French surgeon, Dieulafoy's lesion is a calibre persistent submucosal artery associated with a minute mucosal defect and is a rare but potentially life threatening cause of massive, recurrent GI bleeding. Its reported incidence as a source of upper GI haemorrhage is approximately 2%.[2] It may occur anywhere in the GI tract but it is predominantly located in the gastro-oesophageal junction and in the stomach along the lesser curvature.[3] Dieulafoy'slesion are rarely described in children and the youngest case reported is in an eight week old.[4]The diagnosis may be difficult because of its minute size and intermittent nature of the associated bleeding.

In our case, child presented with hematochezia and clear nasogastric aspirate hence he was evaluated for a presumptive lower GI haemorrhage.[5] In addition, the presentation was complicated by rash. Though Tc 99^{m} scan is the recommended diagnostic technique for Meckel's diverticulum, its reported accuracy is between 85 and 91%.[6] As colonoscopy was not diagnostic, child was subjected to laparotomy. Diverse and predominantly abdominal manifestations including massive lower GI haemorrhage due to mesenteric vasculitis are reported in Henoch-Schönleinpurpura (HSP) especially in younger patients and may be an important differential diagnosis in a child presenting with rash.[7] The rash in this child wasurticarial.Evaluation of a patient with massive GI haemorrhage include selective mesenteric angiography and embolisation, Tc 99^{m} labelled autologous erythrocyte/sulphur colloid scintigraphy and arterial phase multi-detector row helical computer tomography each with its own limitations.[8,9] Age and non availability precluded the use of these techniques .With the clear nasogastric aspirate and predominant lower GI bleed child was not subjected to upper GI endoscopy initially. Though endoscopic procedures are described for the diagnosis and treatment of Dieulafoy's lesion, the pick up rate is around 60% as the bleeding is pulsatile and intermittent.[10] For the same reason, probably the lesion was missed in the first laparotomy.

Upper GI bleed especially duodenalcan present with clear nasogastric aspirate and hematochezia. Vascular malformations should be included in the differential diagnosis of massive gastrointestinal hemorrhage. Upper and lower gastrointestinal endoscopy are indicated prior to laparotomy in a child with gastrointestinal bleeding of undiagnosed etiology.

REFERENCES

1. Oldham KT, Lobe TE.Gastrointestinal hemorrhage in children. A pragmatic update.PediatrClin North Am. 1985 Oct;32(5):1247-63.
2. Avlan D, Nayci A, Altintaş E, Cingi E, Sezgin O, Aksöyek S.An unusual cause for massive upper gastrointestinal bleeding in children: Dieulafoy's lesion.PediatrSurg Int. 2005 May;21(5):417-8.
3. Baettig B, HaeckiW,Lammer F, Jost R. Dieulafoy's disease: endoscopic treatment and follow up. Gut. 1993 Oct;34(10):1418–21.
4. Stockwell JA, Werner HA, Marsano LS. Dieulafoy's lesion in an infant: a rare cause of massive gastrointestinal bleeding. J PediatrGastroenterolNutr. 2000 Jul;31(1):68-70.
5. Kalyoncu D, Urganci N, Cetinkaya F. Etiology of upper gastrointestinal bleeding in young children. Indian J Pediatr. 2009 Sep;76(9):899-901.
6. Tripathi M, Chandrashekar N, Agarwala S, Kumar A, Dasan JB, Malhotra A. Diagnosis of ectopic gastric mucosa using 99Tcm-pertechnetate: spectrum of scintigraphic findings. Br J Radiol. 2005 Aug;78(932):714-20.
7. Ebert EC, Kumar R. Gastrointestinal manifestations of Henoch-SchonleinPurpura. Dig Dis Sci. 2008 Aug;53(8):2011-9.
8. Howarth DM. The role of nuclear medicine in the detection of acute gastrointestinal bleeding. Semin Nucl Med. 2006 Apr;36(2):133-46.
9. Yoon W, Jeong YY, Shin SS, Lim HS, Song SG, Jang NG, Kim JK, Kang HK. Acute massive gastrointestinal bleeding: detection and localization with arterial phase multi-detector row helical CT. Radiology. 2006 Apr;239(1):160-7.
10. Stojakov D, Velicković D, Sabljak P, Bjelović M, Ebrahimi K, Spica B, Sljukić V, Pesko P. Dieulafoy's lesion: rare cause of massive upper gastrointestinal bleeding. ActaChirIugosl. 2007;54(1):125-9.

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