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CASE REPORT

Pyogenic granuloma of the ampulla of Vater: unexpected cause of gastrointestinal bleeding

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Abstract

We describe the case of a previously healthy 8-year-old girl presenting with a 1-year history of iron deficiency anemia. There was no report of hematemesis, abdominal pain or melena. Laboratory work-up excluded iron malabsorption as the underlying cause. Therefore, endoscopic evaluation was performed to exclude gastrointestinal blood loss, which revealed the presence of a 7 mm reddish lesion located within the ampulla of Vater. Capsule endoscopy excluded alternative diagnoses and concomitant lesions. Histopathological examination confirmed the diagnosis of pyogenic granuloma. The young age of the child and the benign nature of this lesion along with the absence of complications favored conservative management. Pyogenic granuloma is a benign vascular lesion that presents as a polypoid red mass. In the gastrointestinal tract, it is a rare condition and occurs more commonly in the elderly. The most common sites are the small intestine, esophagus, and colon, but they can occur throughout the entire gastrointestinal tract with a propensity to bleed that may cause iron deficiency anemia. In pediatric age patients, there are few reports of gastrointestinal pyogenic granulomas, most of which occur in the colon and rectum. Its identification and location in the ampulla of Vater is an exceptional finding.

Keywords Ampulla of Vater · Iron deficiency anemia · Gastrointestinal bleeding · Pyogenic granuloma

Introduction

Pyogenic granuloma (PG) (also known as lobular capillary hemangioma) is a benign vascular proliferation that occurs mostly on the skin and in the oral mucosa, which most commonly affects children and young adults. However, PG in the gastrointestinal tract is very rare and often occurs in the elderly [1, 2]. Trauma, irritation, underlying arteriovenous malformations, and the overproduction of angiogenic growth factors have all been proposed to play a potential role in its development. They manifest as a solitary sessile or pedunculated polypoid lesion that is prone to bleeding and ulceration. Indeed, gastrointestinal PG is a rare cause of gastrointestinal bleeding but should be considered in the differential diagnosis of chronic gastrointestinal bleeding and iron deficiency anemia. We report on a child with iron deficiency anemia whose diagnostic work-up indicated the diagnosis of PG located within the ampulla of Vater.

Clinical case

An 8-year-old girl was referred because of a 1-year history of iron deficiency anemia associated with fatigue and pica. She denied hematemesis and abdominal pain, and there was no change in weight, bowel pattern, or stool color. Her medical history was unremarkable, and her physical examination was normal. The laboratory data (complete blood count, biochemistry, and urinalysis) evidenced hypochromic and microcytic anemia with a hemoglobin level of 9.3 g/dL, normal platelet count, and normal coagulation parameters and inflammatory markers. An iron study revealed a serum iron level of 14 µg/dL (reference range 50–150 µg/dL) with a total iron binding capacity of 398 µg/dL (reference range 258–389 µg/dL), and ferritin of 3.0 ng/mL (reference range 2–178 ng/mL).

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The oral iron challenge study with hydroxide-ferric polymaltose excluded iron malabsorption as the underlying cause. Therefore, endoscopic evaluation was performed to exclude gastrointestinal blood loss. Upper endoscopy (EVIS EXERA II GIF-Q180, Olympus, Japan) showed a protruding reddish lesion located within the ampulla of Vater (Fig. 1a, b). Neither red clots nor active bleeding were noted, despite an apparently friable mucosa. Esophageal and gastric mucosa were normal, as was histological evaluation. Total colonoscopy excluded mucosal lesions or vascular anomalies. Subsequently, wireless capsule endoscopy confirmed the presence of the ampullary lesion and it was otherwise normal.

At this point, we believed that this lesion could be the cause of her iron deficiency anemia but further characterization was needed. Therefore, miniprobe endoscopic ultrasonography (mEUS) was performed and showed an ampulla with slightly heterogeneous thickening, isoechogenic with the submucosa but with apparent "millimetric" hypoechoic structures in its interior. The lesion was superficial with thickening of the mucosal layer and a maximum dimension of 7.5 × 6.2 mm (Fig. 2). Contrast-enhanced abdominal computed tomography showed a small enhanced polypoid lesion in the second portion of the duodenum measuring 7.8 mm, which excluded other lesions and structural anomalies. Small biopsy specimens were obtained, and a bleeding tendency was noticeable. Histological examination revealed a dense vascular proliferation of various sizes with acute and chronic inflammation and granulation tissue. These histological features were consistent with those of a PG (Fig. 3).

The young age of the child and the benign nature of this lesion along with the absence of complications favored conservative management. The patient was prescribed oral iron supplementation with laboratory normalization. However, relapse was noted upon iron discontinuation, causing removal of the lesion to be considered.

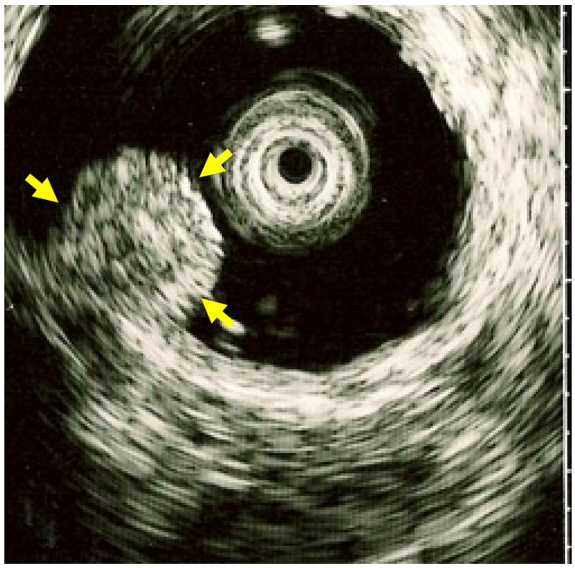

Fig. 2 Miniprobe ultrasound image of the ampulla showing the presence of a superficial lesion (arrows) with involvement and thickening of the mucosal layer, with maximum dimension of 7.5 × 6.2 mm (miniprobe endoscopic ultrasonography, 12 MHz)

Discussion

PG (or lobular capillary hemangioma) is an acquired benign vascular tumor that often develops as a lesion of the skin or oral mucosa. In the gastrointestinal tract, PG is a rare condition and occurs more commonly in the elderly. The most common sites are the small intestine, esophagus, and colon, but they can occur throughout the entire gastrointestinal tract with a propensity to bleed that may cause iron deficiency anemia. In pediatric patients, there are few reports of PG in the gastrointestinal tract, most of which occur in the colon and rectum [3, 4]. The location in the ampulla of Vater, such as occurred in our case, is an exceptional finding raising a diagnostic and therapeutic challenge. In the literature, only three cases of PG in the ampulla of Vater have been reported

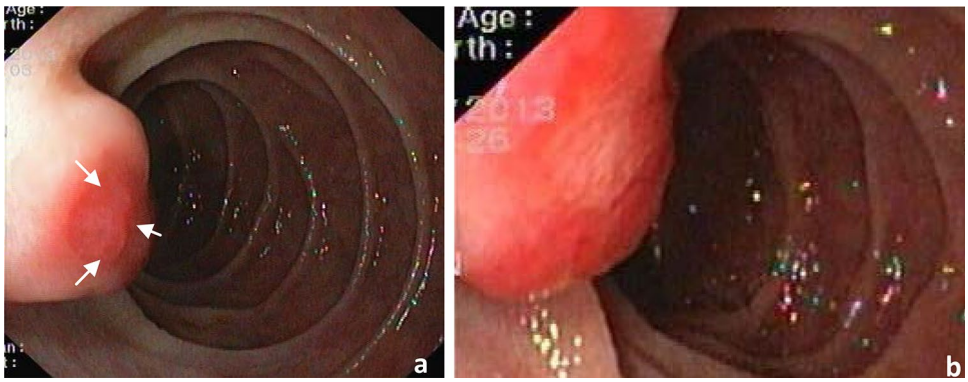

Fig. 1 **a** Endoscopic appearance of the capillary haemangioma (arrows) in the ampulla of Vater (forward viewing endoscopy); **b** close-up of the protruding reddish lesion, appearing friable but with no ulcers or clots

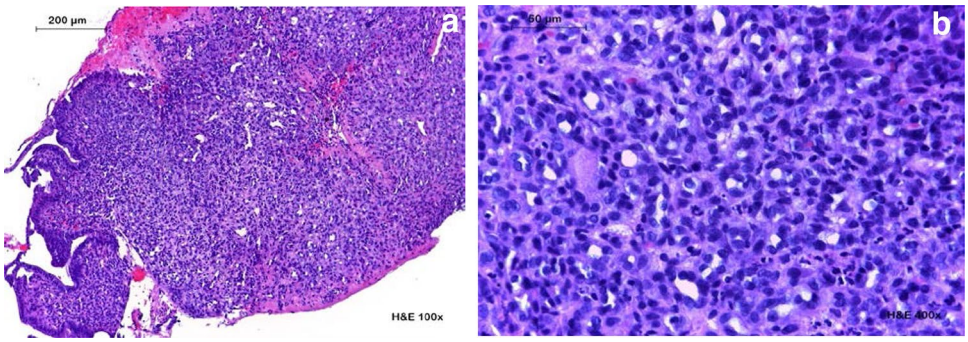

Fig. 3 Histology. **a** haematoxylin and eosin staining, original magnification ×100; **b** haematoxylin and eosin staining, original magnification ×400 showing a proliferation of capillaries of various sizes, lined with endothelial cells and with acute and chronic inflammation

in elderly adults [5–7]. To the best of our knowledge, this is the first reported case in a pediatric patient.

PGs are thought to be a reactive lesion resulting from tissue injury, followed by an impaired wound-healing response with vascular growth. Although mechanical irritation or trauma, infection, and pharmacologic causes have been proposed as etiologic factors, there is still controversy regarding the mechanisms underlying the development of this tumor. In our patient, none of those predisposing conditions were identified, though the underlying pathophysiology in this case remains unclear.

The gross appearance is often characterized by a polypoid form with a smooth surface along with the occasional appearance of ulcers or erosions. They appear erythematous and friable with easy contact bleeding, as their hypervascular nature predisposes them to hemorrhage. The size ranges from a few millimeters to centimeters, and they are generally solitary [5]. The patients usually present with occult gastrointestinal bleeding and anemia. Rarely, these lesions can be associated with overt bleeding. PG has also caused common bile duct obstruction in a single case [8]. The diagnosis can be challenging and often relies on endoscopic techniques and radiologic images. Based on endoscopy, PG is a pedunculated polyp that is mostly red in color with a white coating and occasional ulcerations. In this case, the location in the ampulla of Vater warranted further characterization. Endoscopic ultrasonography usually shows that the lesions are homogeneously hyperechogenic, which reflects the proliferation of blood vessels in lesions such as hemangiomas. Capsule endoscopy is also a valuable diagnostic application, excluding alternative diagnoses and concomitant lesions, mainly jejunal or ileal arteriovenous malformations.

The definitive diagnosis is based on histopathologic evaluation and is characterized by proliferating capillaries with a lobular arrangement. Other features could include inflammation, superficial stromal edema, capillary dilatation, and granulation tissue [2].

Concerning the management of the lesions, endoscopic or surgical resections are often required and are curative. Polypectomy or endoscopy mucosal resection is the optimal treatment for most of the gastrointestinal PGs [9]. In large or deep lesions, a surgical excision may be necessary. However, in the case of major papilla involvement, we believe that treatment options should be individualized and tailored to the characteristics of the patient. Two out of the three cases of PG in the ampulla of Vater reported in the literature underwent endoscopic treatment with good outcomes. Endoscopic ampullectomy followed by argon plasma coagulation (for any residual lesion) after placement of a protective pancreatic duct stent was reported in one patient [7]. On the second patient, the lesion was removed en bloc using an endoscopic snare with coagulation current [5]. In our patient, PG was considered the cause of her iron deficiency anemia. Yet, the benign nature of the lesion along with the absence of overt gastrointestinal bleeding favored conservative management. However, endoscopic resection will be required, as the hypervascular nature predisposes to recurrent hemorrhage. The young age of this patient and location in the ampulla of Vater poses a therapeutic challenge, as it can be more susceptible to complications and incomplete resections are prone to recurrence [1].

In conclusion, PG, although rare, must be considered in the differential diagnosis of iron deficiency anemia in the context of gastrointestinal bleeding, even in pediatric patients.

Author contributions HMS drafted the initial manuscript, critically reviewed the manuscript, and approved the final manuscript as submitted. GS, EC, FP and RL critically reviewed the manuscript and approved the final manuscript as submitted.

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