CLINICAL CASE

Solitary hemangioma of the small bowel disclosed by wireless capsule endoscopy

Hémangiome unique de l’intestin grêle révélé par vidéocapsule endoscopique


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Summary  A nine-year-old child presented with melena and anemia. She had similar symptoms five months earlier and had undergone an extensive workup with upper gastrointestinal endoscopy and colonoscopy, both normal and 99m-Tc-RBC-scintigraphy which was positive in the right lower quadrant. This time, capsule endoscopy was performed and disclosed an hemangioma with a dark spot suggesting recent bleeding in the ileum. The lesion was resected. Pathological examination revealed a transmural cavernous hemangioma. Small bowel hemangioma is a rare disease. Its diagnosis is extremely difficult and is usually obtained during surgery. Capsule endoscopy is an endoscopic technique that can improve preoperative diagnosis, as reported in the present case.

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Résumé  Les hémangiomes de l’intestin grêle sont rares et ont longtemps été de diagnostic difficile. Nous rapportons l’observation d’une fillette de neuf ans qui présentait un mélèna et une anémie. Cinq mois auparavant une gastroscopie et une coloscopie, qui étaient normales, avaient déjà été réalisées, ainsi qu’une scintigraphie aux hématies marquées qui était positive au niveau du quadrant inférieur droit. Lors de la deuxième hospitalisation, un examen par vidéocapsule endoscopique révélait un hémangiome de l’intestin grêle centré d’une image évoquant un caillot suggérant une hémorragie récente. Une intervention chirurgicale permettait la résection de la lésion qui à l’histologie, se révélait être effectivement un hémangiome caverneux.

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Introduction

Hemangioma of the small bowel is a rare disease that usually presents as gastrointestinal bleeding. Preoperative diagnosis, which is extremely difficult, is classically performed through angiography, small bowel series, push enteroscopy or scintigraphy. The majority of cases were, therefore, only diagnosed during surgery. The preferred treatment is surgical when isolated lesions are found. The authors report a case of small bowel hemangioma diagnosed through wireless capsule endoscopy, currently the preferred technique for mucosal imaging of the small bowel.

Case report

A nine-year-old child presented to our emergency department with fatigue, dizziness and nausea. She had past history of growth retardation since five months old. Physical examination only revealed cutaneous-mucous pallor. Laboratory exams revealed anemia (7.6 g/dl) (normal range: 11.5—15.5) with normal mean corpuscular volume (85.9 fl) (normal range: 77—95) and mean corpuscular hemoglobin concentration (31.9 g/dl) (normal range: 31—37) and iron deficiency (serum iron: 17 μg/dl [normal range: 37—158], total iron binding capacity: 271 μg/dl [normal range: 228—428], transferrin saturation: 6.3%, ferritin: 8.51 ng/ml [normal range: 13.0—150.0]). She was admitted for symptomatic anemia. During admission, melena with further hemoglobin decrease to 5.6 g/dl occurred. Continuing clinical and analytical improvement was observed, after blood transfusion. Upper gastrointestinal endoscopy and colonoscopy were normal. Scintigraphy with 99 mTc-labeled red cells was positive in the right lower quadrant (Fig. 1).

She was discharged on oral iron replacement therapy and maintained regular outpatient follow-up. Five months later, melena and anemia (9.8 g/dl) (normal range: 11.5—15.5) recurred. Upper gastrointestinal endoscopy was again normal. Wireless capsule endoscopy was scheduled three days later, after interruption of iron replacement therapy and overnight fast. A bluish polypoid mass (Fig. 2.1) without active bleeding but with a dark spot (Fig. 2.2) suggesting an hemangioma with recent bleeding was observed in the ileum. The lesion was referred to the distal ileum, since the capsule reached the cecum a few minutes later. Surgical treatment was proposed. A bluish-purple vascular lesion averaging 2.5 cm was observed 80 cm from the ileocecal valve. The involved bowel segment was resected (Fig. 3). Pathological examination revealed a 2 × 2.5 cm transmural cavernous hemangioma, eroding the mucosa and extending to the serosa (Fig. 4). Postoperative follow-up was uneventful and she remained asymptomatic without further blood loss.

Figure 1 99 mTc-RBC-scintigraphy. Positive in the right lower quadrant.

Scintigraphie aux globules rouges marqués au 99 mmTc, fixation au niveau de l’hypocondre droite.

Discussion

Small bowel hemangioma is a rare disease accounting for 7—10% of benign tumors of the small intestine [1]. These tumors may be solitary or multiple and are more frequent in the jejunum [2,3]. Enteric hemangiomas are more common when present as components of the blue rubber-bleb nevus syndrome [4], Maffucci’s syndrome [5], Klippel—Trenaunay syndrome [6] or disseminated neonatal hemangiomatosis [7]. No manifestations of these syndromes were present in our patient.

Microscopically, hemangiomas may be classified in cavernous, capillary or mixed types [8]. The most common is cavernous hemangioma, the type found in our patient. They are described as large blood-filled lakes or blood-filled sinuses lined by endothelium, separated by a connective tissue matrix. Grossly, they are blue, purple or red-colored soft polypoid lesions, sessile or pedunculated, varying in size from a few millimeters to several centimeters [1].

The usual presentation is intestinal bleeding usually insidious presenting as anemia or acute and potentially life-threatening [3]. Other forms of presentation, albeit rare, have been described including intussusception [9], obstruction or perforation [10].

The diagnosis is very difficult and usually requires an extensive workup. In a review of the Japanese literature, preoperative diagnosis was obtained in only 24.1% of patients [11]. Several exams have been used to diagnose small bowel hemangiomas, all with significant limitations, including plain films, small bowel follow-through, upper gastrointestinal endoscopy, push-enteroscopy, angiography, scintigraphy, CT and MRI [1,3,12].

WCE is a technique that revolutionized the approach of patients with obscure gastrointestinal bleeding. It is perfor-
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Median increasingly earlier in the diagnostic workup, replacing other tests like red cell scintigraphy that was still performed in our patient without providing additional diagnostic information. A report was recently published in which WCE located the source of bleeding in the ileum in a patient bleeding from an ileal hemangioma [13]. However, the diagnosis was only made during surgery through intra-operative enteroscopy. Other similar case reports have been published recently, in which capsule endoscopy allowed the diagnosis of a solitary small bowel hemangioma [14,15]. The relevance of WCE in the diagnosis of small bowel lesions has been extensively verified. A recent series of five small bowel tumors diagnosed by WCE, among which one jejunal capillary hemangioma and one hemangiosarcoma has been reported [16].

WCE is generally well tolerated in children above nine years old and has the same yield found in adult patients. The main concern is to ascertain the patient’s ability to swallow the capsule and to assess the age and weight that will permit the capsule to pass through the pylorus and ileocecal valve. It has been safely used in children with age above five years old and weight above 17 Kg [17].

Intra-operative enteroscopy will continue to be useful, in some hemangiomas not identified from the serosal side during surgery. In these cases, intra-operative enteroscopy is important to define the location and extent of the lesions [3].

Surgery was until recently the only curative treatment available. In cases of multiple hemangiomas or when complex lesions are found [10], curative surgery is impracticable. Double balloon enteroscopy, a new endoscopic technique in the exploration and treatment of small bowel diseases has again changed the approach of these lesions. A

Figure 2 Capsule endoscopic images. Fig. 2.1 Hemangioma in the small bowel at 2 h 21 min. Fig. 2.2 Hemangioma with dark spot (red arrow).

Images fournies par la capsuloscopie. Fig. 2.1 Hémangiome de l’intestin grêle à deux heures et 21 minutes. Fig. 2.2 Hémangiome avec une tache noire.

Figure 3 Surgical specimen with hemangioma. Prélèvement chirurgical montrant l’hémangiome.

Figure 4 Histology: H & E, low magnification. Blood-filled pools separated by connective tissue, below the epithelial layer. Histologie : H & E, faible grossissement. Plages remplies de sang séparées par des travées de tissu conjonctif sous la couche épithéiale.
case report was recently published [18] in which a patient with blue rubber nevus syndrome with digestive tract involvement and severe iron deficiency anemia was successfully treated with double balloon enteroscopy. Small bowel hemangiomas were removed through polypectomy and argon plasma coagulation without complications. Available medical treatment options include steroids, embolization and alpha-interferon, with variable results [10].

**Conclusion**

Small bowel hemangioma, like other forms of obscure gastrointestinal bleeding, have been a diagnostic and therapeutic challenge over the years. Recently, small bowel endoscopy, either through wireless capsule endoscopy or through double balloon enteroscopy have completely changed the diagnostic workup of small bowel lesions. They dramatically increase the odds of an early and accurate diagnosis and, in several cases, provide the opportunity of noninvasive therapy. The diagnosis of small bowel hemangioma has also changed. Small bowel hemangioma will be increasingly diagnosed before surgery, mostly through capsule endoscopy and, in some cases, double balloon enteroscopy will be used for therapy.

**References**