Capsule Endoscopy Diagnosis of Celiac Disease and Ileal Tumors in a Patient with Melena of Obscure Origin

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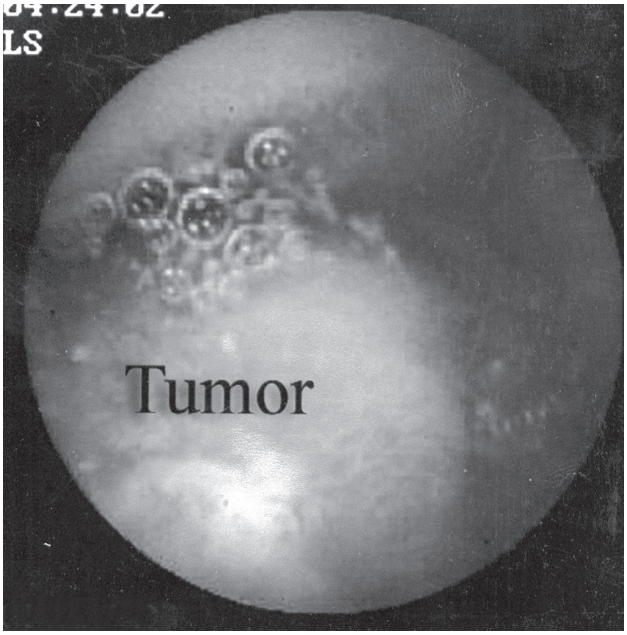
Obscure gastrointestinal bleeding is defined when conventional endoscopies (esophagogastroduodenoscopy and colonoscopy) fail to show a potential source of the bleeding. Obscure GI bleeding can be overt (e.g., melena) or occult (e.g., chronic iron deficiency anemia). The source of obscure bleeding is usually located in the small bowel. Most of the small bowel is beyond the reach of the push enteroscope. The diagnostic yield of radiographic imaging is usually low in this setting [1]. The PillCam capsule endoscopy system (Given Imaging Ltd., Yokneam, Israel) provides direct non-invasive visualization of the entire small bowel [2]. We report the case of an elderly man with melena of unknown origin in whom capsule endoscopy diagnosed small bowel tumors and an edematous mucosa.

Patient Description

A 74 year old man was hospitalized with a 24 hour history of melena and an increased number of stools/day during the preceding few weeks. His past history was unremarkable, except for splenectomy for splenic hydatid cyst 17 years before. Apart from pallor and tachycardia, his physical examination was normal. Routine laboratory tests revealed a hemoglobin level of 7 g/dl and mild hypoalbuminemia (3.2 g/L). Esophagogastroduodenoscopy, colonoscopy and abdominal computed tomography scan were normal. CE demonstrated a diffusely edematous mucosa along the duodenum and the jejunum, and two ileal tumors (1–2 cm in size), one of which was ulcerated [Figure].

GI = gastrointestinal
CE = capsule endoscopy

Enteroscopy revealed the edematous mucosa. Biopsies showed mucosal atrophy with characteristics compatible with celiac disease, undiagnosed previously. Later, serologic tests showed an elevated level of anti-gliadin immunoglobulin G (33 EU/ml, normal 0–20 EU/ml), a borderline level of anti-endomesial antibody, and a high level of tissue-transglutaminase antibody (>150 EU/ml, normal 0–20 EU/ml). Laparotomy findings were similar to those of CE. Two limited resections were performed. Surgical pathology showed a carcinoid tumor (10–12 mm in size) with low mitotic activity in each of the two resection specimens, and a gastrointestinal stromal tumor, 1 cm in size, with benign features, located near one of the carcinoid tumors.



An ileal ulcerated carcinoid tumor photographed by capsule endoscopy 4 hours 24 minutes after the patient swallowed the capsule.

Comment

Small bowel tumors are relatively rare. Two-thirds of them are malignant, representing only 1.1%–2.4% of gastrointestinal malignancies. The prevalence of small bowel tumors among patients undergoing CE is relatively low. In 27 studies (of consecutive patients) published or presented at international meetings, a tumor was found in 34 (3.8%) out of a total of 892 patients, including 772 patients with obscure overt and obscure occult bleeding.

In the sole published series on small bowel tumors diagnosed by capsule endoscopy, four tumors (two stromal tumors, one hemangiosarcoma and one capillary hemangioma) were diagnosed among 130 patients with obscure bleeding. The fifth tumor (a carcinoid 1 cm in diameter) was diagnosed in a patient with abdominal pain. In three of the five patients an enteroclysis was performed, which was not diagnostic [3].

Most of the lesions disclosed by CE in cases of obscure bleeding are non-tumoral (e.g., angiodysplasia). Obviously, the diagnostic yield of conventional

small bowel imaging is very low in these patients [1]. The diagnostic accuracy of small bowel follow-through radiography in cases of small bowel tumors (with heterogeneous clinical presentation) is only 30–44%. In one retrospective radiologic study, enteroclysis showed a tumor in 18 of 20 patients, including relatively small carcinoids. However, the diagnostic yield of this procedure is operator-related and there is a lack of further information in the literature regarding this yield. Studies in the future will probably show that CE is superior to radiologic modalities in diagnosing small bowel tumors and will permit earlier diagnosis. Although the clinical importance of small tumors discovered by CE is not clear (unless they bleed during the examination), in our patient one of the carcinoid tumors was ulcerated and was therefore probably the source of the bleeding. We should also remember that in the small bowel, even small tumors like small carcinoids are potentially malignant.

We believe that in cases of obscure bleeding, CE should be the first diagnostic step after negative results with conventional endoscopies. When CE results are negative or when the findings are only suspicious (e.g., a single angiodysplasia), an enteroscopy should be performed. Conversely, if a tumor is initially disclosed by push enteroscopy or suspected on radiologic imaging, CE should be performed in order to clarify the diagnosis or to exclude synchronous lesions. For example, about one-quarter of carcinoid tumors are multiple, as in our patient. For these reasons and owing to the limitations of CE, it is worthwhile performing an intraoperative enteroscopy before surgical excision of a small bowel tumor.

Apart from ileal tumors, CE revealed an edematous mucosa of the duodenum and the jejunum in our patient. Subsequently, previously unknown celiac disease was diagnosed by serology and histology. A new diagnosis of celiac disease is occasionally made even during the eighth decade, usually in patients with iron deficiency anemia. The predisposition of celiac patients to small bowel lymphomas and adenocarcinomas is well established. However, only three case reports of small intestinal carcinoid [4] have been published, only one case report of a small intestinal leiomyoma and no cases of small intestinal GI stromal tumor, among patients with celiac disease. Among 395 patients with primary small bowel malignancies diagnosed in the UK during a 2 year period (including 175 adenocarcinomas and 107 lymphomas), 65 suffered from celiac disease. All the 79 carcinoids were among the non-celiac patients [5]. Therefore, we believe that the concurrent diagnosis of celiac disease and ileal carcinoids in our patient was coincidental.

In summary, we report on a patient with melena of unknown origin in whom CE diagnosed small bowel tumors and an edematous mucosa due to celiac disease which was previously unknown. This report emphasizes the role of CE in patients with obscure GI bleeding.

References

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