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CASE REPORT

Heterotopic Mesenteric Ossification Following Gastric Bypass Surgery: Case Series and Review of Literature

Arthur Yushuva · Prachi Nagda · Kei Suzuki · Omar H. Llaguna · Dimitrios Avgerinos · Elliot Goodman

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Abstract Heterotopic mesenteric ossification (HMO) is a rare entity with few cases reported in the world literature. We report two cases. Both patients underwent an open gastric bypass with Roux-en-Y reconstruction procedure for morbid obesity and subsequently presented with gastrointestinal fistulae associated with HMO.

Keywords Heterotopic mesenteric ossification · Ectopic bone formation · Rectus abdominis muscle flap

Introduction

Heterotopic mesenteric ossification (HMO) is a rare intra-abdominal bone-producing pseudosarcoma typically associated with abdominal trauma, previous laparotomy, neoplasia, or intra-abdominal infection. HMO patients are typically middle-aged to elderly men with a prior history of abdominal surgery [1, 2]. Our two patients demonstrate many of the distinctive clinical features of HMO.

A. Yushuva (✉) · P. Nagda · K. Suzuki · O. H. Llaguna · D. Avgerinos · E. Goodman
Department of Surgery, Beth Israel Medical Center,
Albert Einstein College of Medicine,
First Avenue at 16th Street,
New York, NY 10003, USA
e-mail: ayushuva@yahoo.com

Case Reports

Case 1

The first patient is a 50-year-old male with morbid obesity (body mass index [BMI]=57) and past medical history of hypertension, diabetes, hypercholesterolemia, spinal stenosis, and bipolar disorder who underwent gastric bypass and cholecystectomy. His postoperative upper gastrointestinal (UGI) series was negative for leak and the patient was discharged home on postoperative day 2. The patient presented to the emergency department on postoperative day 12 with milky discharge from the superior portion of his incision but without signs of sepsis. A repeat UGI at this time showed an enterocutaneous (EC) fistula, likely in the Roux limb of the jejunum. A computed tomography (CT) scan of the abdomen with oral and intravenous contrast showed linear densities in the mesentery (Fig. 1).

The patient was taken to the operating room for exploratory laparotomy. At exploration, an enterotomy was found in the midportion of the Roux limb, but of note was the development of severe fibrosis and thickening of the small bowel mesentery, greater omentum, and peritoneum, which had encased the entire small bowel just below the EC fistula, causing distal obstruction. Multiple biopsies showed fat necrosis with prominent reactive fibrosis and ossification and no evidence of atypical cells or mitotic figures making a rare pathologic diagnosis of HMO (Fig. 2).

During the re-exploration, a small bowel resection was performed in an attempt to control the fistula; however, due to the severe fibrosis of the Roux limb, the anastomosis broke down. The patient was taken back to the operating

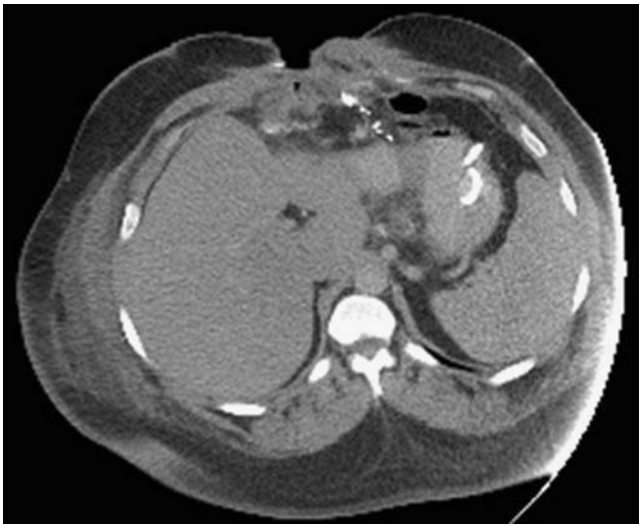

Fig. 1 CT scan of the abdomen with oral and intravenous contrast showing linear radiopaque densities in the mesentery

room 5 days later for further exploration. At re-exploration this time, a small leak was noted from the staple line. Since the small bowel was noted to be very edematous, a decision was made to intubate the leak rather than to primarily repair it and the skin was left open. The open abdominal wound and the EC fistula were eventually controlled with a vacuum-assisted closure device, and the patient was discharged home.

The patient developed multiple recurrent EC fistulae over the period of a year, requiring repeated hospital admissions, small bowel resections, and fistula repairs. Definitive closure of the fistula ultimately required correction with a rectus abdominis muscle flap. The flap closure was successful and the patient is now symptom free for the past 6 months.

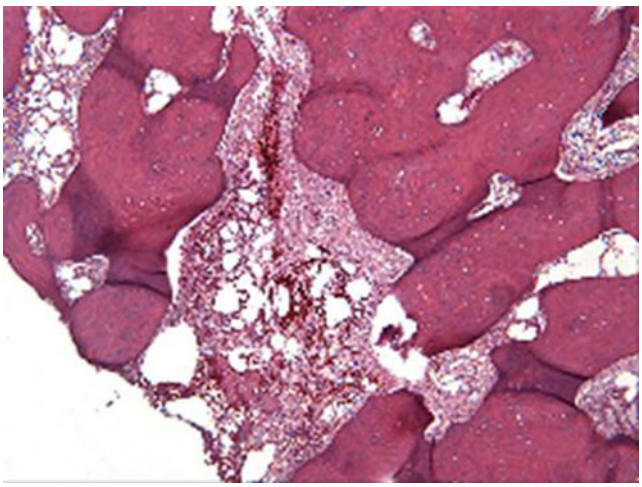

Fig. 2 Mesenteric biopsy showing fat necrosis with prominent reactive fibrosis and ossification

Case 2

The second patient is a 42-year-old male with past medical history of morbid obesity (BMI=44), sleep apnea, and hypothyroidism who underwent Roux-en-Y gastric bypass. After discharge, the patient presented to the emergency room with signs of an anastomotic leak, at which time he was taken back to the operating room for repair of a gastrojejunal leak and placement of feeding jejunostomy tube. A Jackson–Pratt drain was placed at surgery. The leak, however, persisted, and the patient developed a chronic gastrocutaneous fistula. The patient was readmitted on several occasions, and several unsuccessful attempts were made to repair the fistula endoscopically with local application of fibrin glue.

The patient presented for a second opinion, at which time the fistula was approximately 10 weeks old. The fistula drained 10–30 ml/day of murky fluid. Giving the presence of a mature tract of granulation tissue lining the fistula, the Jackson–Pratt drain was removed. The fistula initially healed but then recurred.

The patient was seen in clinic 10 months after the initial surgery, at which time a freely draining subcutaneous abscess was noted at the site of the old gastrocutaneous fistula. A CT scan showed a 5×6-cm mass just below the costal margin in between the midline and the fistulous tract in the left upper quadrant. This was associated with air trapping from the Roux limb of the jejunum consistent with a new EC fistula (Fig. 3). The patient's white blood cell count at the time was 14,000/mm^3; however, the patient

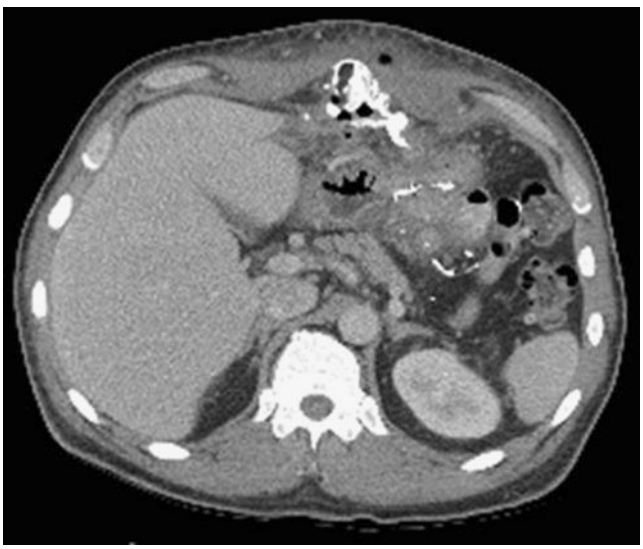

Fig. 3 CT scan of the abdomen with oral and intravenous contrast showing curvilinear radiopaque mesenteric densities in the upper anterior abdominal wall

was afebrile. He presented to the emergency room 2 days later with complaints of malaise and erythema overlying the skin.

The patient was taken back to the operating room for exploratory laparotomy, revision of gastric bypass, and gastrostomy tube placement. Intraoperatively, a 6×8-cm hard abdominal mass was identified near the midline, within which some bone fragments were noted. The mass was excised. Pathologic examination of the mass showed fibroadipose soft tissue with acute and chronic inflammation with reparative changes and prominent ossification; there was no evidence of cellular atypia or mitotic figures making the diagnosis of HMO (Fig. 4).

The patient's postoperative course was complicated by a *Klebsiella* wound infection: the same organism had grown out of the abscess prior to surgery. He was discharged home on postoperative day 8. He was seen in clinic after surgery and is currently doing well.

Discussion

HMO is a distinct intra-abdominal ossifying pseudotumor, typically occurring in males, almost always after surgery or abdominal trauma. It frequently presents with symptoms of intestinal obstruction or, as in our cases, recurrent EC fistula [2]. The exact cause and effect relationship of fistula formation and HMO in our two patients remains undetermined. Our first patient had HMO developed distal to the fistula tract, whereas our second patient's HMO was closely related to the fistula. Nevertheless, it is generally accepted that HMO is an exuberant reactive process in response to various stimuli such as mechanical and surgical trauma, venous stasis, ischemia, edema, inflammation, and local osteoinductive factors (e.g., bone morphogenetic protein) [3]. It is an inflammatory reaction with transformation of fibroblast or adipose tissues in bone cells [5]. The presence of clearly reactive zones resembling nodular fasciitis and thick osteoid and the absence of nuclear atypia, necrosis, and atypical mitotic figures allow the distinction of HMO from its most important morphologic imitate, extraskeletal osteosarcoma [2]. The lesions of HMO usually develop after one or more intra-abdominal surgical procedures (although the literature reports a few patients without prior history of surgery) and tend to grow rapidly, developing in days or weeks after the stressor [3].

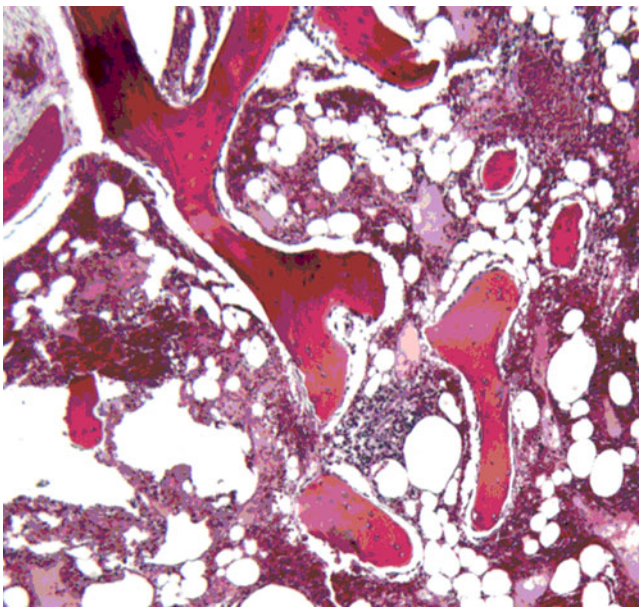

Fig. 4 Small bowel mesentery showing fibroadipose tissue with extensive ossification

The preoperative diagnosis of HMO is difficult, although a trabecular architecture as seen on radiographic studies is highly suggestive of ossification. A CT scan may provide confirmatory evidence [1]. Radiologically, differentiation between HMO and dystrophic calcification, osseous neoplasia, or oral contrast leakage remains difficult. Ossification is characterized by a trabecular pattern, whereas calcification appears as irregular, punctate, and faint radiodense areas that do not possess trabecular or cortical structure. Furthermore, the presence of trabecular bone differentiates HMO from ossifying neoplasms, which produce poorly organized bone. Finally, it can be distinguished from oral contrast leakage because the latter will not remain unchanged on subsequent images, should collect in the most dependent point, and is not trabeculated [4].

Surgical excision may be needed because of complications such as intestinal obstruction and, as in our cases, recurrent EC fistulas. Local radiotherapy or treatment with diphosphonates or anti-inflammatory agents has been shown to decrease recurrence in orthopedic literature [6]. We recommend against such therapy in bariatric patients. Radiation causes intestinal wall fibrosis and ischemia leading to ulceration and anastomotic leaks [7]. Although not shown in any trials, nonsteroidal anti-inflammatory drugs may contribute to mucosal ulceration via cyclooxygenase inhibition and loss of prostaglandin-mediated mucosal barrier [8]. Use of diphosphonates is contraindicated due to malabsorption of calcium after an intestinal bypass and vitamin D deficiency that is frequently seen in gastric bypass patients [9].

In conclusion, HMO is a rare entity with unknown etiology and can be easily misdiagnosed. Radiological work up is usually helpful in the diagnosis. Treatment is primarily surgical excision.

Conflict of Interest The authors declare that they have no conflict of interest.

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