Spontaneous heterotopic mesenteric ossification around the pancreas causing duodenal stenosis: A case report with literature review

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ABSTRACT

INTRODUCTION AND IMPORTANCE: Heterotopic mesenteric ossification (HMO) is a rare condition that can be hereditary or nonhereditary. It can lead to small bowel obstruction, which may require corrective surgery. Most affected patients have a history of abdominal surgery or trauma. Spontaneously occurring HMO is even rarer, with only 7 cases reported till date. There has been no previous report of spontaneous peripancreatic HMO.

CASE PRESENTATION: A 60-year-old man presented with complaints of recurrent nausea and vomiting for 2 months. Esophagogastroduodenoscopy revealed luminal stenosis and edematous changes involving the second and third parts of the duodenum but not its complete obstruction. Abdominepelvic computed tomography showed faintly enhanced thickening of the involved duodenal walls along with mild dilatation of the common bile duct. Considering the possibility of peripanillary cancer, we performed a pylorus-preserving pancreatoduodenectomy. Histopathological examination confirmed the diagnosis of HMO with extensive fibrosis involving the peripancreatic soft tissue.

CLINICAL DISCUSSION: The peripancreatic HMO with severe fibrosis can occur duodenal stenosis, and it is mimicking peripanillary cancer. However, the preoperative diagnosis of spontaneous HMO is difficult, and a diagnosis confirmed after surgery.

CONCLUSION: Herein, we described our experience of managing a rare case of duodenal stenosis due to spontaneous HMO involving peripancreatic tissue.© 2021 The Authors. Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY license (http://creativecommons.org/licenses/by/4.0/).

1. Introduction

Heterotopic mesenteric ossification (HMO) is defined as the development of a bony lesion within the intra-abdominal mesentery [1–3]. There are <40 cases of HMO reported in the literature [3]. However, a spontaneous presentation is extremely rare, and there have been no reported cases of HMO surrounding the pancreas. Here, we describe a rare case of a patient diagnosed with spontaneous HMO involving peripancreatic tissue, a presentation mimicking that of peripanillary cancer. This work has been reported in line with the Surgical Case Reports guidelines [4].

2. Presentation of case

A 60-year-old man presented with complaints of recurrent nausea and vomiting for 2 months. No history of abdominal surgery or trauma was noted. There were no specific abnormalities in the laboratory analyses, carcinoembryonic antigen (CEA) and carbohydrate antigen 19-9 (CA19-9). Abdominal X-ray indicated a mildly distended stomach.

We inserted a nasogastric tube and drained 50–300 ml of bile-stained gastric fluid for 3 days. After the stomach was fully decompressed, we performed an esophagogastroduodenoscopy (EGD), which showed both a large amount of bile fluid and food material in the stomach. The EGD further revealed luminal stenosis and edematous changes affecting the second and third parts of the duodenum, though the involved lumen was not obstructed completely (Fig. 1A). Computed tomography (CT) images of the patient’s abdomen and pelvis showed faintly enhanced thickening of the involved duodenal walls along with mild dilatation of the common bile duct (Fig. 1B).

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Considering the possibility of cancer of either the pancreatic head or the periampullary region, we performed an exploratory laparotomy. Intraoperatively, we observed massive fibrosis and adhesions surrounding the second and third parts of the duodenum and the head of the pancreas. The entire pancreatic body was hardened, and the connective tissue around the head of the pancreas showed severe desmoplastic changes. We performed sophisticated adhesiolysis to release the duodenum along with a pylorus-preserving pancreaticoduodenectomy.

The histopathology report confirmed the diagnosis of heterotopic ossification with extensive fibrosis of peripancreatic soft tissue. Grossly, we identified an ill-demarcated, dark-red-to-tan, soft, and fleshy lesion in the periampullary region (Fig. 2A). Microscopic examination revealed calcified lesions admixed with fibrous and adipose tissue, within the peripancreatic soft tissue. However, the metaplastic bone deposits did not show any atypia (Fig. 2B,C). The postoperative course was uneventful, and the patient was discharged 2 weeks after the operation.

3. Discussion

This report highlights the case of a patient with spontaneous HMO, which is an extremely rare disease presentation that can involve the periampullary region and have diagnostic features that mimic those of pancreatic cancer.

Heterotopic ossification (HO) is defined as the formation of bone in non-skeletal tissues and classified into 2 subgroups—hereditary and nonhereditary [1]. Nonhereditary HO (NHHO) is usually associated with trauma, tissue injuries, infection, or surgery [1,5,6]. In
1989, Mirra defined the occurrence of HO in soft tissues as “myositis ossificans” [7]. Intra-abdominal HO was first described by Wilson et al. in 1999 [2]. Since then, there have been a few reports of HMO, but most affected patients had a history of trauma, infection, or prior surgery. Only 7 cases of spontaneous HMO have been described in the literature to date (Table 1) [2,8–11]. Although there are various theories regarding the pathophysiological mechanisms of heterotopic bone formation in NHMO, the etiopathogenesis of spontaneous HMO remains unclear [1,12].

It is difficult to diagnose HMO preoperatively. However, detecting trabecular architecture and dystrophic calcifications on a CT scan may provide confirmatory evidence of ossification [9,13]. While 2 of the reported cases involved patients, who were incidentally diagnosed with high-density lesions on CT scans, we found no suspiciously ossified lesions in the imaging studies in this case. HMO patients are known to present with symptoms of small bowel obstruction. While 5 of the previously reported cases involved patients presenting with abdominal symptoms due to small bowel obstruction, our patient experienced recurrent nausea and vomiting caused by the stenosis of the third part of the duodenum. The duodenal stenosis was associated with severe fibrosis, forming a mass-like lesion around the uncinate process of the pancreas. Thus, the appearance of the lesion mimicked that of pancreatic head cancer, a diagnosis suggested by results of both EGD and CT. However, histopathological examination revealed severe inflammation and HO of the peri pancreatic region.

In HMO, surgical management is recommended, and recurrence is rare. As there are a few reports on the effectiveness of additional therapy for prevention of recurrence after surgery, further research is indicated in this regard [1,3,12].

### Table 1
Summary of reported cases of spontaneous heterotopic mesenteric ossification in existing literature.

| Reference (year) | Age (years) | Sex | Trauma/operation | Clinical presentation | Diagnostic procedure |
|------------------|-------------|-----|------------------|-----------------------|----------------------|
| Wilson et al. [2] (1999) | 43 | Male | None | Small bowel obstruction | Surgery |
| Comperat et al. [8] (2004) | 80 | Male | None | Cholelithiasis | Surgery |
| Bosker et al. [9] (2004) | 64 | Male | None | Abdominal mass | Surgery |
| Bovo et al. [10] (2004) | 76 | Female | None | Right flank pain | Surgery |
| Present case (2020) | 70 | Male | None | Small bowel obstruction | Surgery |
| Choi et al. [11] (2008) | 69 | Male | None | Abdominal mass | Needle biopsy |
| Present case (2020) | 60 | Male | None | Duodenal stenosis | Surgery |

### Consent
Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor of this journal.

### Author contribution
Conceptualization: Hanlim Choi, Dong Hee Ryu.
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Writing – review & editing: Jae-Woon Choi, Dong Hee Ryu, Chang Gok Woo.

### Registration of research studies
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### Guarantor
The guarantor is Jae-Woon Choi.

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