European Journal of Radiology Open 8 (2021) 100318

Contents lists available at ScienceDirect

European Journal of Radiology Open

journal homepage: www.elsevier.com/locate/ejro

Ectopic pancreas with lipoma in the distal jejunum: A case report

Ji-Eun Kim

Department of Radiology, Seongnam Citizens Medical Center, #171, Sujeong-ro, Sujeong-gu, Seongnam-si, Gyeonggi-do, Republic of Korea

ARTICLE INFO

Keywords:
Ectopic pancreas
Lipoma
Jejunum
Submucosal tumor
Computed tomography

ABSTRACT

Ectopic pancreas is asymptomatic and mostly found in the stomach and proximal small bowel. It is difficult to distinguish ectopic pancreas with other submucosal tumors of the gastrointestinal (GI) tract on computed tomography (CT) without histological confirmation. We encountered a case of ectopic pancreas coexisting with submucosal lipoma in the distal jejunum which was misdiagnosed as fat-containing submucosal tumor. We report the CT findings and review previously reported cases of ectopic pancreas coexisting with submucosal lipoma. The knowledge that ectopic pancreas can occur with benign lipoma, and its imaging features would be helpful for the diagnosis and management of patients.

1. Introduction

Ectopic pancreas is the presence of histologically-confirmed pancreatic tissue outside the anatomic boundaries of the pancreas and lacks both anatomical and vascular connections [1]. Cases of ectopic pancreas have been identified throughout the entire length of the gastrointestinal (GI) tract, but the majority of the cases occur in the stomach and the proximal small bowel [2]. Computed tomography (CT) findings of ectopic pancreas is nonspecific; therefore, diagnosis before pathological-confirmation is difficult. Moreover, the differential diagnosis of ectopic pancreas occurring in the GI tract and coexisting with other tumors might be challenging. There are a few reported cases of ectopic pancreas coexisting with other tumors, which were mostly malignant or premalignant lesions [3]. On the contrary, the present case of ectopic pancreas coexisted with benign lipoma, which was misdiagnosed as a fat containing submucosal tumor on CT scan. Herein, we report a case of ectopic pancreas with lipoma in the distal jejunum.

2. Case presentation

A 38-year-old man visited our institution for evaluation of melena, intermittent epigastric and left upper quadrant pain lasting for one week. He had no medication history of non-steroidal anti-inflammatory drugs (NSAIDs) or aspirin. On physical examination, he had abdominal tenderness in epigastric area and the bowel sounds were normal. Digital rectal examination showed remnants of melena without other pathological findings. He got the Levin-tube irrigation of stomach and clear fluid was presented. At the time of admission, his systolic/diastolic blood pressure was 95/45 mmHg. Laboratory examination revealed the hemoglobin level of 6.1 g/dL and the hematocrit level of 18.3 %. A contrast-enhanced computed tomography (CT) scan of the abdomen and pelvis revealed a 3.2 cm sized lobulated contour soft tissue mass in the distal jejunum. The mass was intraluminally protruded and overlying mucosa was intact. At the proximal portion, the mass was connected with homogeneous fat density portion (Fig. 1A). The possibility of submucosal tumor containing fat component, such as liposarcoma or submucosal tumor coexisting with lipoma was suggested. There was no evidence of small bowel obstruction nor ischemic change. During surgery, small bowel mass with intussusception was found, and laparoscopic assisted segmental resection of small intestine and anastomosis was performed. On the gross examination, the specimen showed an elongated polypoid mass ("finger-like" appearance) in a segment of the distal jejunum. The mass was measured as 8.0 cm × 2.0 cm × 1.5 cm. The adjacent mucosa was grossly normal (Fig. 1B). Microscopically, the polypoid lesion was covered by the mucosa. In the submucosa, groups of pancreatic tissue (pancreatic acini, islets of Langerhans, and pancreatic ducts) were present. The pancreatic tissue was closely associated with a different tumor composed of mature fibro-adipose tissue (Fig. 1C). It suggests ectopic pancreas with co-occurred lipoma. After surgery, the patient was discharged in a few days without complications.

3. Discussion

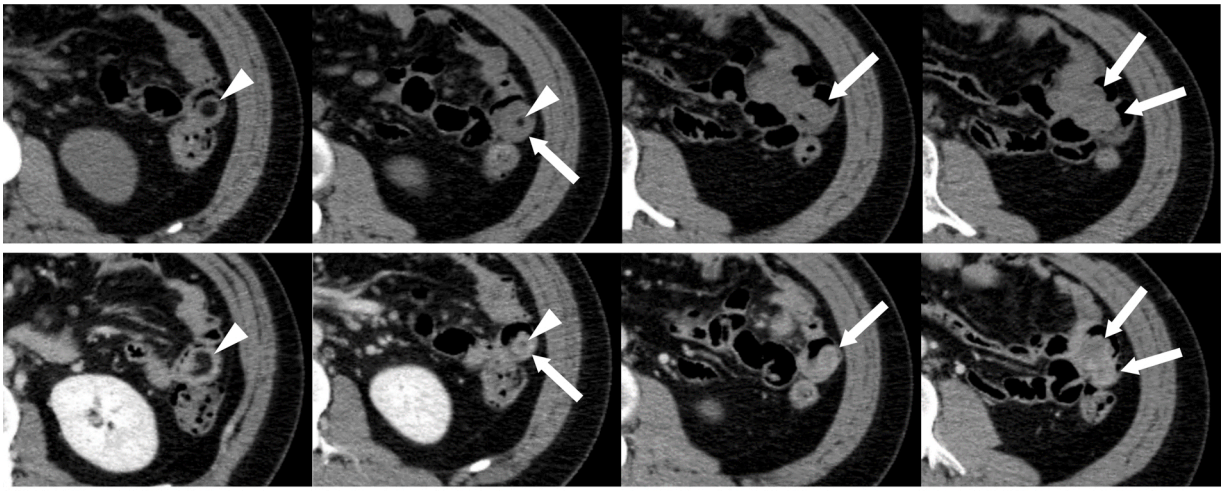
Ectopic pancreas is a congenital anomaly in which the pancreatic tissue separates from the main gland and exists without vascular or ductal continuity [1]. Lipomas are benign tumors composed of mature

E-mail address: dieun22@nate.com.

https://doi.org/10.1016/j.ejro.2020.100318
Received 27 August 2020; Received in revised form 19 December 2020; Accepted 21 December 2020

fat tissue [4]. Radiological findings of ectopic pancreas are similar to those of normal pancreas, and often present as ectopic nodular foci [5]. However, ectopic pancreas demonstrates nonspecific CT density and enhancement pattern, similar to other submucosal tumors [6]. On the contrary, gastrointestinal lipoma may appear as an intraluminal mass with gross appearance similar to that of subcutaneous fat on CT [4].

In our case, the mass showed both submucosal enhancing soft tissue and fat component. Therefore, the differential diagnosis was difficult with the possibility of fat-containing submucosal tumor, such as submucosal tumor coexisting with lipoma and liposarcoma. Meanwhile, all the prior cases and present case of ectopic pancreas associated with lipoma were diagnosed following complications, likely related to the presence of lipoma itself rather than ectopic pancreas [2,7–11]. Therefore, in relation to the clinical relevance, simple lipoma may be the



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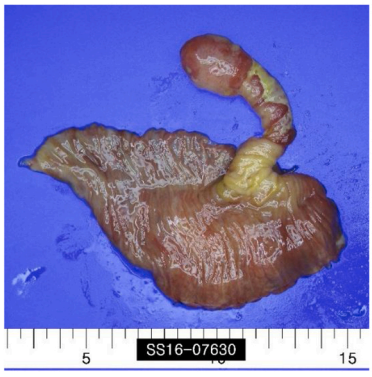

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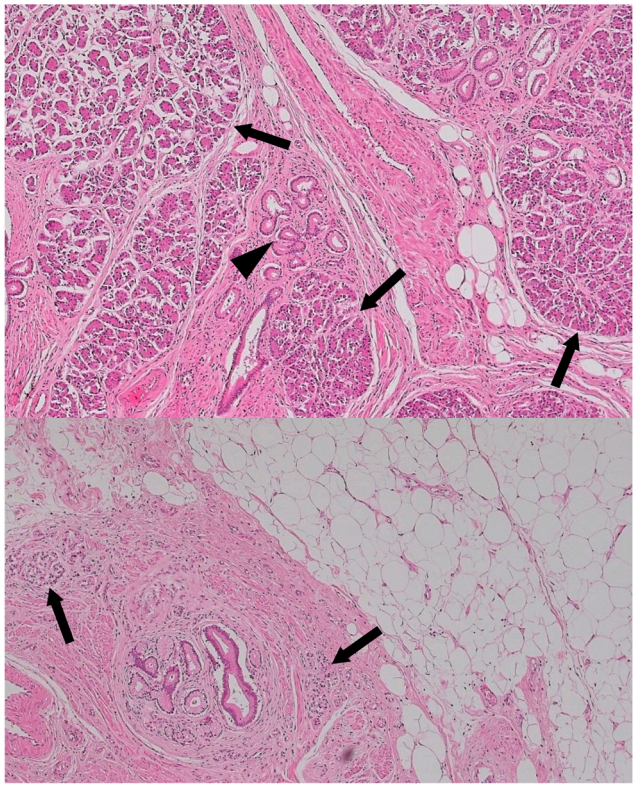

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Fig. 1. A 38-year-old male with ectopic pancreas with lipoma in the distal jejunum.
A. Serial axial precontrast (the upper row) and contrast-enhanced (the lower row) CT images reveal ectopic pancreas coexisting with lipoma in the distal jejunum. A 3.2 cm sized lobulated contoured enhancing soft tissue mass **(arrows)** in the distal jejunum. It is intraluminally protruded and overlying mucosa is intact. At the proximal segment of the lesion, the enhancing soft tissue mass is connected with homogeneous fat density portion **(arrowheads)**.
B. Gross specimen shows the elongated polypoid mass ("finger-like" appearance) in a segment of the distal jejunum measuring 8.0 cm × 2.0 cm × 1.5 cm.
C. Microscopic specimens of the mass in the distal jejunum (H&E stain, ×40).
The upper panel shows pancreatic tissue with acini **(arrows)** and ducts **(arrowhead)**.
The lower panel shows lipoma tissue composed of mature fibro-adipose tissue at the boundary of pancreatic tissue. The arrows indicate islets of Langerhans.

following preoperative radiological differential diagnosis. Next to the simple lipoma, the differential diagnoses of ectopic pancreas with submucosal lipoma include several types of tumors. Liposarcomas have various CT patterns based on the amount and distribution of fat in the tumor, and often present as fatty masses with enhanced soft-tissue density [12]. Gastrointestinal stromal tumor (GIST) can present as an enhanced intraluminal mass or extend through the serosa as an exophytic mass. Leiomyoma is typically a round, well-circumscribed mass with soft-tissue density with homogeneous enhancement and frequent ulceration and bleeding, as in the present case [6]. Neuroendocrine tumors can appear as polypoid, plaque-like, or hyperenhancing mass and up to 70 % of the cases show calcification. Moreover, presence of metastases and multiple primary tumors in other organs and findings of stranding on CT because of fibrosis and desmoplastic reaction are common in case of mesenteric metastases [6].

Retrospective examination of the CT findings of our case showed that the lipomatous region of the mass was clearly distinct from the ectopic pancreas and grossly appeared as fat, which corresponded to the imaging findings of lipoma in the GI tract. The observations suggest the possibility of two different origins for the tumor coexisting in the same location.

Each of these two conditions is well-known but rarely reported together in the same location in the GI tract. Previously reported cases of ectopic pancreas coexisting with tumors were malignant or premalignant lesions. For example, Tu et al. reported 26 cases of heterotopic pancreas coexisting with digestive tract tumors; 23 cases of adenocarcinoma (20 cases in the stomach and 3 cases in the colon), 2 cases of gastrointestinal stromal tumor (1 case each in the stomach and the proximal jejunum) and duodenal hemangioma [3].

On the contrary, only 6 cases of ectopic pancreas coexisting with benign lipoma have been reported (Table 1). Although ectopic pancreas has been reported to occur throughout the entire length of the GI tract, the majority of the cases occur in the stomach and proximal small bowel. Lipoma can occur anywhere along the gastrointestinal tract as well. The most common site for bowel lipoma is the colon (65–75 %), followed by the small bowel (20–25 %) and the stomach (5 %) [4]. Ectopic pancreas with submucosal lipoma in previously reported cases were mostly located in the ileum (6 cases), and in the distal jejunum in our case, and may have a tendency to occur in the distal small bowel. Contrary to the fact that most patients with ectopic pancreas are asymptomatic, all reported patients with ectopic pancreas coexisting with submucosal lipoma had symptoms and complications, such as GI bleeding, intussusception, or bowel obstruction, including in this case [2,7–11]. It is likely to be caused by the presence of lipoma rather than ectopic pancreas. Therefore, it is important to suggest the differential diagnosis of simple lipoma, especially for possible cases in relation with lipoma and encountered incidentally on abdominal examination performed due to other indications, to prevent complications.

The precise etiology of each ectopic pancreas and lipoma remains unclear. Ectopic pancreas is considered to be caused by abnormal implantation of fragments of the ventral or dorsal pancreatic anlage in the intestinal wall during embryological development. Other theories, such as pancreatic metaplasia of endodermal tissue, have been proposed to explain ectopic pancreas in distant organs [13]. Lipomas are benign with limited information on their pathogenesis. An increased incidence of lipoma is noted with obesity, diabetes mellitus, high serum cholesterol, trauma, irradiation and familial history [14]. However, no predisposing factor was identified for the present patient.

Limited options are available for the confirmative diagnosis and treatment of the small bowel lesions. Given the location of the lesion and presence of possible complications, such as gastrointestinal bleeding, obstruction, or intussusception, conventional or laparoscopic resection with anastomosis is considered an appropriate treatment till date for ectopic pancreas and lipoma in distal small intestine with good prognosis [13].

Despite the difficulty associated with preoperative radiological diagnosis of ectopic pancreas with coexisting lipoma, knowledge of the fact that ectopic pancreas can occur with benign lipoma and its imaging features would be helpful for the diagnosis and management of patients.

Table 1
Previously reported cases and present case of ectopic pancreas with lipoma.

No.	Authors	Reported year	Sex/ Age	Location	Clinical presentation
1	Fikatas et al. [2]	2008	M/ 28	Distal ileum	GI bleeding
2	Chuang et al. [11]	2010	F/26	Distal ileum	Ileoileal intussusception, Bowel obstruction
3	Jiang et al. [7]	2015	F/38	Distal ileum	GI bleeding, Ileoileal intussusception, Bowel obstruction
4	Musthafa et al. [10]	2015	M/ 32	Mid ileum	GI bleeding, Ileoileal intussusception, Bowel obstruction
5	Sunitha et al. [8]	2017	M/ 35	Ileum	Ileoileal intussusception, Bowel obstruction
6	Papageorge et al. [9]	2018	M/ 36	Terminal ileum	Ileoileal intussusception, Bowel obstruction
7	Present case	2019	M/ 38	Distal jejunum	GI bleeding

4. Conclusion

Ectopic pancreas coexisting with lipoma is a rare presentation and can be misdiagnosed with other submucosal tumors of the GI tract on CT scan. Knowledge of the co-occurrence of ectopic pancreas with benign lipoma and its imaging features will help radiologists and clinicians to make appropriate therapeutic and management decisions before opting for invasive procedures.

Ethical statement

There is no ethical problem in this article to be declared.

Funding statement

The author has no disclosure of financial interest to report.

Declaration of Competing Interest

The authors report no declarations of interest.

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