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Review Article

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AN UNEXPECTED DIAGNOSIS OF ECTOPIC PANCREAS IN THE LIVER

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ABSTRACT

A 54-year-old hypertensive female patient underwent surgical resection of the sigmoid colon two years ago for a moderately differentiated adenocarcinoma classified as pT3N1b. During the follow-up period, CT scan disclosed multiple metastases predominant in the right hepatic lobe, hence the initiation of neoadjuvant chemotherapy (4 courses FOLFOX - ERBITUX). The patient underwent a right hepatic lobectomy. Histological examination of the different samples taken from the liver confirmed the presence of a secondary hepatic localization of a moderately differentiated adenocarcinoma of colorectal origin with healthy limits of excision. In the surrounding liver there was macrovacuolar steatosis estimated at 20%. In addition, we noted a focus of pancreatic heterotopia including acini and excretory ducts organized in a lobule. The final pathological diagnosis was that of pancreatic heterotopia type II according to Heinrich classification. Postoperative course was uneventful. During the follow-up period, the patient developed liver metastases in the left hepatic lobe. The diagnosis of heterotopic pancreas prior to surgery is difficult due to its non-specific clinical signs and symptoms. Diagnosis can only be established based on histopathological examination.

KEYWORDS: Ectopic pancreas, liver, pathology.

INTRODUCTION

The ectopic pancreas or pancreatic heterotopia is defined by the presence of pancreatic tissue in an abnormal situation, with no anatomical relationship to the main gland. The usual locations of ectopic pancreas are the duodenum (30-35%), the stomach (30%) and the jejunum (15%).[1] Pancreatic heterotopia is rarely located in the liver. We herein report a new case of an incidentally discovered ectopic pancreas in the liver.

CLINICAL HISTORY

A 54-year-old hypertensive female patient underwent surgical resection of the sigmoid colon two years ago for a moderately differentiated adenocarcinoma classified as pT3N1b. During the follow-up period, CT scan disclosed multiple metastases predominant in the right hepatic lobe, hence the initiation of neoadjuvant chemotherapy (4 courses FOLFOX - ERBITUX). The patient underwent a right hepatic lobectomy. Histological examination of the different samples taken from the liver confirmed the presence of a secondary hepatic localization of a moderately differentiated adenocarcinoma of colorectal origin with healthy limits of excision. In the surrounding liver there was macrovacuolar steatosis estimated at 20%. In addition, we noted a focus of pancreatic heterotopia including acini and excretory ducts organized in a lobule. This focus measured approximately 2 mm. The final pathological diagnosis was that of pancreatic heterotopia in the liver classified as Heinrich's type II. Postoperative course was uneventful. During the follow-up period, the patient developed liver metastases in the left hepatic lobe.

Table 1: Reported cases of ectopic pancreas in the liver.

Case	Author (year)	Age/sex	Symptoms	Associated Diseases	Heinrich classification
1	Ballinger J [3] (1941)	53, M	Hypoglycemia	Insulinoma (pancreas)	I
2	Mobini J [4] (1974)	68, F	Abdominal tumor	None	II
3	Lieutaud R [5] (1978)	39, M	None	Seminoma	II
4	Payan MJ [6] (1985)	46, M	Epigastralgia Retrosternal pain	Caroli disease	II
5	Nishiguchi Y [7] (1988)	42, M	Jaundice	Choledocolithiasis	II
6	Terada T [8] (1990)	41 cases (autopsy) 18M/23F	-	-	II (100%)
Our case	Limaiem F (2020)	54, F	Abdominal pain	Colorectal cancer Liver	II

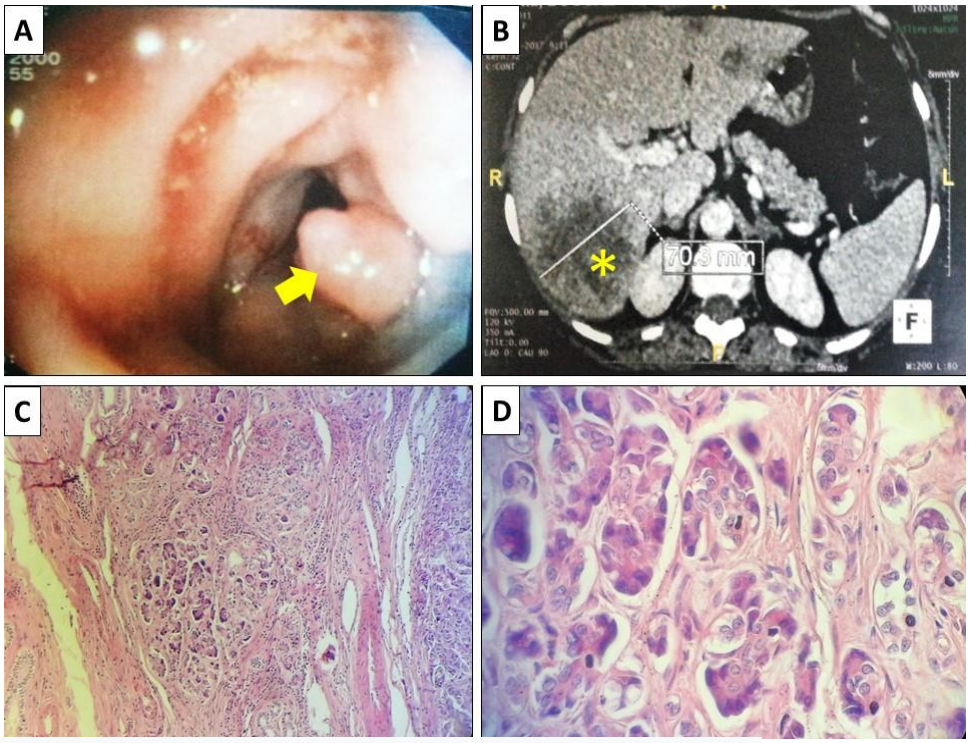

Figure 1A: Endoscopic appearance of the tumor of the sigmoid colon (arrow).
Figure 1B: Abdominal computed tomography showing multiple hepatic nodules predominant in the right lateral sector of size varying between 0.6 and 7 cm (asterisk).
Figure 1C: Ectopic pancreatic lobule in the hepatic parenchyma. (Hematoxylin and eosin, x 200) (Arrow).
Figure 1D: Pancreatic acini and excretory ducts devoid of cytonuclear atypia. (Hematoxylin and eosin, × 400).

DISCUSSION

Ectopic pancreas (also referred as pancreatic heterotopia, pancreatic choristoma, accessory pancreas, adenomyoma, aberrant pancreas, or pancreatic rest) is a congenital anomaly lacking vascular or anatomic continuity with the main body of the pancreas.[1,2] The most common locations of ectopic pancreas include the upper gastrointestinal tract mainly, the stomach, duodenum, and proximal jejunum.[1,2] Heterotopic pancreas in the liver is very rare with only 46 cases reported in literature (Table 1).[3-8] The mean age of patients with ectopic pancreas in the liver is 50.16 years (range: 1 month – 84 years) with a slight female predominance: sex ratio (M/F) = 0,916.[3-8] In one study, heterotopic pancreas in the liver was found in 41 (4.1 %) of the 1000 livers. The high incidence of ectopic pancreas in the liver noted in this study may be explained by the fact that adequate tissue sampling was performed.[8] The origin of heterotopic pancreas is controversial. Several theories have been advanced to explain ectopic pancreas, however, none is universally accepted. In one study, the authors found intrahepatic heterotopic pancreas only in the large portal tracts, and there was no ectopic pancreas in the small portal tracts or in the hepatic lobules. The development of hepatocytes, biliary tract, and pancreas are closely related. The liver and biliary tract arise from the embryonic foregut as a hepatobiliary bud. From its proximal part, the ventral pancreatic bud develops. Therefore, it is possible that certain portions of the ventral pancreatic bud detach, are incorporated into hepatobiliary anlagen, and eventually migrate into the large portal tracts to form intrahepatic

heterotopic pancreas.[8] Uncomplicated heterotopic pancreas is typically asymptomatic, and is usually incidentally discovered during an unrelated surgery as it was the case in our patient, or during an imaging examination, or at autopsy.[8] It is difficult to diagnose hepatobiliary heterotopic pancreas preoperatively, and the definitive diagnosis relies on histopathological examination.

Heinrich established the classification of ectopic pancreas in 1909. Three types of heterotopic pancreas are described in this classification:
Type I: includes ducts, acini, and endocrine islet cells.
Type II: consists of ducts, acini and without islet cells.
Type III: is exclusively composed of pancreatic ducts.[9,10] In our case, ectopic pancreas included acini and ducts without islet cells thus corresponding to Heinrich's type II. Among the 46 ectopic pancreas reported in literature, 45 were classified as type II and only one case was classified as type I according to Heinrich classification[3-8] (Table 1). Heterotopic pancreatic tissue is susceptible to the same pathologic conditions that can affect the orthotopic pancreas, including pancreatitis, pseudocyst formation, and malignant transformation. None of the 46 cases of intrahepatic ectopic pancreas presented such complications.

In conclusion, we present the case of an incidental finding of an intrahepatic ectopic pancreas in a 54-year-old woman with a past medical history of sigmoid adenocarcinoma metastatic to the liver. Preoperative diagnosis of heterotopic pancreas in the liver is very difficult especially in case of very small lesions as it was the case of this patient. The mainstay of diagnosis is histological.

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