



A Case of Invasive Adenocarcinoma Arising from Heterotopic Pancreas in the Stomach and Review of the Literature

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Abstract

Heterotopic pancreas is a rare condition of tissue found outside the pancreas without anatomic connection with ectopic one. Generally asymptomatic and incidentally findings could be located elsewhere in the gastrointestinal tract. Degeneration into neoplasm is rare. Less than sixteen cases has been describe. Our case is the first well reported an adenocarcinoma arising from a locally advanced stomach's HP discovered for hematemesis.

Introduction

The Heterotopic Pancreas (HP) is defined as a pancreatic tissue found outside the eutopic pancreas without anatomic or vascular connections between them.

It is not uncommon with an incidence of approximately 0.5% - 13.7% in autopsy studies and 0.5% in upper abdominal laparotomies and It can be found anywhere in the gastrointestinal tract. The most common site is the stomach, mostly in the antrum and prepyloric region on the greater curvature or posterior wall. The heterotopic pancreas is usually an incidental finding, however, mass-like manifestations causing pyloric obstruction, ulcer, and bleeding can be observed in the gastric area [1].

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We report a case of a patient with HP of the stomach who developed an invasive adenocarcinoma presenting with hematemesis.

Case Report

A 76 years old woman, with a past medical history of hypertension, under evaluation for anemia, was admitted to the Emergency Department (ED) with melena and hematemesis. Hemoglobin was 6.2 g/dl, red blood cell 2.3×10^6 , Hematocrit 22%, elevated lactate and Base Deficit (BD). Her blood pressure was 75 over 55 mmHg and 120 heart pulse rate, euthermic, skin pale and clammy. Coagulation screening was within normal value. She received fluids and blood units. She subsequently underwent Oesophago-Gastro-Duodenoscopy (OGD) that revealed the presence of a blood clot occupying the body of the stomach. During the admission she became unstable so an angio-Computed Tomography (CT) was performed. That showed a neoplastic lesion involving the body of the stomach, transverse colon, distal pancreas and spleen (Figure 1). Due to presence of multiple collateral vessels feeding the lesion with high risk of massive necrosis embolization was not performed. A gastrectomy, spleno-pancreatectomy and resection of the transverse colon with Roux and Y esophagojejunostomy and end to end colo-colic anastomosis was performed.

In the first post-operative period the patient developed Acute Respiratory Distress Syndrome (ARDS) and required external ventilation and hemodynamic support in intensive care unit. The histology revealed/showed adenocarcinoma (pTN4N2M0) (Figure 2 and 3). After 24 days the patient was discharged and referred to the oncological department. Due to comorbidity she wasn't deemed fit for any oncological therapy and, after one year, she died for the progression of her disease.

Discussion

HP is often and incidental finding and many patients with HP are asymptomatic and require



Figure 1: CT scan of the neoplasm arising from EP in stomach (white arrow).

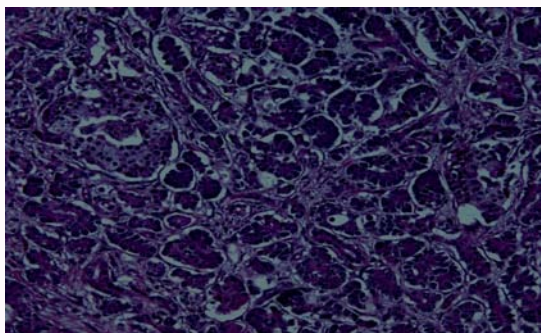


Figure 2: Haematoxylin and Eosin 100X. Ectopic pancreas.

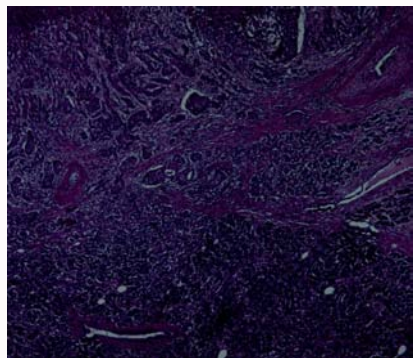


Figure 3: Haematoxylin and Eosin 50X. Passage from HP (in the lower portion of the image) and neoplasm (in the upper part of the image).

no treatment [2]. The rate of HP during autopsy varies from 0.6% to 15% and about 75% of ectopic pancreases are found in the stomach, duodenum, or ileum [3]. In the stomach, HP is most often located along the greater curve of the prepyloric antrum and especially within 6 cm of the pyloric ring. HP are usually less than 4 cm in size, and a central mucosal depression is often recognized at endoscopy. An ectopic pancreas usually invades the submucosal or muscle layer, and has histologically normal pancreatic tissue [4].

Histology confirmation and diagnosis that a malignant tumor has raised from pre-existing ectopic pancreatic tissue may be rather difficult. Described three criteria to prove that a malignancy arose from an ectopic pancreas: 1) the tumor must be within or near the ectopic pancreatic tissue; 2) a direct transition between pancreatic structures and carcinoma must be observed; 3) the non-neoplastic pancreatic tissue must, at a minimum, be fully developed [5].

Heinrich classified the heterotopic pancreas into 3 types [6]: type I, all the components of the pancreas including ducts, acini, and

Table 1: Lists of article concerning Neoplasm arising from HP in the stomach.

Authors	N Pt	Sex	Site	Age
Tanaka K et al. [1]	1	F	Antrum	56
Hickman DM et al. [3]	1	M	Body	60
Jeong H Y et al. [4]	1	M	Antrum	64
Jeong HY et al. [4]	2			
Emerson L et al. [4]	1	M	Antrum	56
Von Heinrich et al. [5]	1	M	Antrum	35
Pang LC et al. [7]	1	F	Body	75
Cho SJ et al. [8]	1	F	Antrum	76
Kim JY et al. [9]	1	M	Pylor	73
Shaib YH et al. [10]	1	M	Antrum	64
Li-Xin Jiang et al. [11]	1	F	Body	57
Rodriguez FJ et al. [12]	1	M	Pylor	58
Gurocak B et al. [13]	1	M	Antrum	80

endocrine islets; type II, ducts with acini; and type III, ducts with a few acini or dilated ducts only, so called adenomyoma. When the pancreaticobiliary-type ducts predominate, they are often surrounded by hypertrophic smooth muscle bundles.

In a series of 32 cases of symptomatic HP none of the patients was diagnosed preoperatively [7]. Barium swallow studies may show a rounded filling defect, sometimes with central indentation. Imaging studies with endoscopic ultrasound (EUS) and (CT) are frequently used for the diagnosis of gastrointestinal submucosal tumor and can be helpful in the diagnosis of gastric HP, but are not specific [8]. However, studies have suggested that both EUS and CT help to distinguish HP from other submucosal tumors. Endoscopic appearance of HP is that of a well-circumscribed submucosal mass with a normal overlying mucosa and a central dimpling which corresponds with the opening of a duct. The characteristic dimpling or umbilication is observed in less than half of the cases, and therefore, HP may easily be misinterpreted as another submucosal tumor such as a stromal tumor, or leiomyoma at endoscopic examination. Because GISTs are by far the most common gastric submucosal tumors, HP can frequently be mistaken for GIST at endoscopy as happened in our case [9]. Endoscopic biopsy performed by using standard biopsy forceps is most often unremarkable. However, a few reported cases of HP were diagnosed with biopsies obtained with the jumbo forceps [10]. Diagnosis can occasionally be made through endoscopic biopsies but those are inconclusive in about 50% of the cases as normal gastric mucosa could cover the HP [11].

EUS-guided aspiration has been reported to be helpful to diagnose HP [12]. If an endoscopic resection is considered, EUS is also extremely useful for pre-excision assessment. There was no correlation between the histological type of HP and the presence of symptoms. Surgery is frequently needed to make a definitive diagnosis and to plan further treatment because the differential diagnosis includes leiomyoma, lymphoma, carcinoid tumors, and other malignancies.

If HP is diagnosed as an incidental finding, local excision is recommended. Symptomatic HP should be resected. Endoscopic excision can be considered in select cases depending on the size and location of the mass, especially for treating the benign lesions of HP.

Because the preoperative imaging studies (ultrasonography,

endoscopic ultrasonography and computerized tomography) are not very specific [13], intraoperative frozen section studies are useful to confirm the diagnosis and exclude the most common differential diagnosis of HP such as gastrointestinal stromal tumor, gastrointestinal autonomic nerve tumor, lymphoma, carcinoid tumor and other malignancies. Recently, the combination of endoscopic ultrasonography with fine needle aspiration cytology from submucosal lesions is showing encouraging results due to its sensitivity which ranges from 80% to 100% [14].

The histogenesis of HP tissue is controversial. Yamagiwa et al. [15] suggested that Heinrich type I HP of the stomach was caused by migration from fetal pancreas, while Heinrich type II and III heterotopic pancreas arises through erroneous differentiation of primitive gastric mucosal epithelium. The real incidence of HP into the stomach is uncertain; in a large series [15,16]. They examined the whole stomach by numerous sections in gastrectomies. They showed 107 cases of heterotopic pancreas out of 5,446 surgically resected stomachs; the incidence was 1.2%. Reported 12.7% incidence of cancer in their series of HP in the 1980 [17].

The review of the literature, showed 14 cases well documented of neoplasm arising from pancreatic ectopia in the stomach (Table 1). The site most involved is the antrum and generally the lesion was well confined into in stomach wall. Surgery ranged from small resection to total gastrectomy. Overall survival was good. No case of diffuse cancer as our case has been described in the past. The majority of carcinomas arising with in HP are adenocarcinoma sora plastic carcinomas. Review of the literature reveals adenocarcinomas arising within heterotopic pancreas to have a better prognosis than patients with adenocarcinoma of the pancreas.

In conclusion, based on the literature the findings of HP is not so uncommon. The gastric localization of HP is most common among the number of HP, but, based on literature's data, more than ten cases developed cancer. The correct approach for diagnosis includes endoscopic ultrasound with several biopsies, fine needle aspiration and CT scan. A resection remains the gold standard if HP is suspected.

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