Case Report

Heterotopic pancreas in a chronic calculous cholecystis - A rare case report

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ABSTRACT

Context: Heterotopia of the pancreas is the presence of pancreatic tissue in an abnormal location having no anatomical or vascular continuity with the main body of the pancreas which is a rare entity. It is a congenital condition and takes years for heterotopic pancreas to become symptomatic. The presentation of heterotopic pancreas with cholecystitis is very rare with its clinical significance remain unclear so far.

Case report A 58 year old female patient presented with the history of abdominal pain since 3 days. Abdominal ultrasonography revealed features suggestive of acute calculous cholecystitis. The patient was hospitalized and laparoscopic cholecystectomy was performed. Gross examination showed thickened wall with dull mucosa and ulcerated areas at places. Multiple tiny stones were also noted. Microscopy showed ulcerated mucosa with transmural infiltration with chronic inflammatory cell. Submucosa showed pancreatic tissue comprised mainly of ductal and acinar components. This case of pancreatic heterotopia in the gall bladder is worth reported because of its rarity.

Key Word- Heterotopic pancreas, gall bladder, cholecystitis, choristoma, cholecystectomy

INTRODUCTION

Heterotopia of the pancreas, also called pancreatic choristoma, is defined as pancreatic tissue in an abnormal location with no vascular, neuronal or anatomical continuity with the main body of the pancreas. Heterotopic pancreas is very rare and found commonly in the stomach, duodenum, proximal jejunum and also in Meckel's diverticulum. At times, it is also rarely found in the ileum, bile ducts, gallbladder, splenic hilum, umbilicus and liver. Heterotopic pancreas in the upper GIT accounts for 90% amongst all the reported cases.^{1,2} In the majority of cases, the heterotopic pancreas is an incidental finding following a cholecystectomy performed for symptomatic gallbladder disease.,3

In this report, we present a case report of heterotopic pancreas in gallbladder with clinical findings of acute cholecystitis.

CASE REPORT

A 58 year old female presented with pain abdomen since 3 days which was aggravated on movement. On physical examination, the right hypochondrium was tender with a positive Murphy's sign, but there was no rebound tenderness. Abdominal ultrasound revealed features suggestive of acute calculous cholecystitis. Biochemical, serological and hematological findings were normal except for the total leucocyte count which was 23,400 cells/cu mm with 90% of neutrophils. Laparoscopic cholecystectomy was carried with preoperative diagnosis of calculous acute cholecystitis.

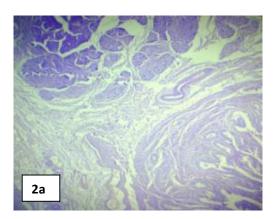
On macroscopic examination, the gallbladder was partially cut opened and measured 6.5x4.5x0.5cm.Wall was thickened at places (body); mucosa was ulcerated and appeared dull. Cut section of the thickened areas was brown.

Multiple tiny pigmented stones were identified with largest measuring 0.5 x0.5cms.



FIGURE 1: Cut opened cholecystectomy specimen received displaying congested serosa and irregular mucosal surface.

Microscopic examination showed ulcerated mucosa with transmural infiltration with chronic inflammatory cell. Submucosa showed aberrant pancreatic tissue consisting of acini, intra and interlobular ducts.



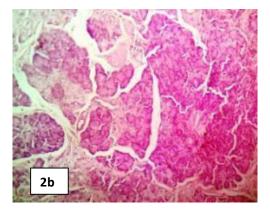


FIGURE 2:(a) Micropicture showing the mucosa and the submucosa with aberrant pancreatic tissue consisting of acini, intra and interlobular ducts. (b) Closer view of the aberrant pancreatic tissue

DISCUSSION

The reported incidence of heterotopic pancreas in the gastrointestinal tract ranges from 0.5% to 13.7% in autopsy series, and 0.2% in laparotomy. Heterotopic pancreas can affect all ages with men are affected usually three times more than women.

But the incidence of heterotopic pancreas of the gallbladder cases in female patients is higher.⁴ Heterotopic pancreas in the gallbladder presents as an exophytic growth and may be similar to a polypoidal lesion or as yellow-colored nodules with variation in dimension from a few millimeters to 4 cm. It arises in the neck of the gallbladder in fifty

percent (50%) of the cases. In our case, heterotopic pancreas was found located at the submucosa from the section taken from the thickened area on the body which is most common presentation and found in 73% of cases. Our histopathological examination revealed a heterotopic pancreas made up of exocrine acinar and ductal components without islet cells, corresponding to incomplete

heterotopia. Only in the one-third of cases, Islets of Langerhans are identified Microscopically, ectopic pancreas has been classified into three types by von Heinrich⁵ (Table 1). Our case was considered to be a type 2 ectopic pancreas, based on Heinrich classification.

Table 1. Heinrich classification of ectopic pancreas	
Type 1	Ectopic tissue with acini, ducts and islets of Langerhans.
Type 2	Ectopic tissue with incomplete or lobular arrangement (only a few acini and multiple ducts). Endocrine elements are absent.
Type 3	Ectopic tissue of proliferating ducts (so-called adenomyoma). Both exocrine acini and endocrine elements are lacking.

Most patients with heterotopic pancreas are asymptomatic, and the heterotopic tissue is found incidentally at histological exam following a cholecystectomy as in our case. Heterotopic pancreas in the gallbladder is rarely symptomatic. The manifestation of symptoms is largely associated with acute or chronic cholecystitis with or without gallstones. There are a few cases reported in which the cause of acute symptoms was due to the inflammation of the heterotopic pancreatic tissue, resembling acute pancreatitis.⁶ The mechanism in which this aberrant tissue producing various symptoms is unclear. It may be suggested that active pancreatic enzymes, such as amylase and trypsin, refluxing into the biliary tract and gallbladder lumen could produce inflammation, spasm and biliary symptoms in patients without gallstones, and hence acute cholecystitis in patients with gallstones.⁷ In addition, heterotopic pancreatic tissue can undergo pathologic changes, such as cyst or abscess formation and calcification. Malignant

transformation of a heterotopic pancreas may occasionally occur. In this case, the tiny pigmented stones found have caused the gallbladder symptomatology in adjunct with the heterotopic pancreas due to the continuous secretion of active pancreatic enzymes, mainly amylase into the gallbladder lumen, causing further damage to the epithelium and producing symptoms associated with chronic inflammation.

The preoperative diagnosis of an aberrant pancreas in the gallbladder is impossible using the imaging techniques currently available. Ultrasonography and computed tomography cannot distinguish an aberrant pancreas in the gallbladder from other lesions, such as cholesterol polyps, adenoma and carcinoma.⁸

And also in the cases of acalculous lesions, such as polypoid formations, isolated parietal thickenings or nodulations, differential diagnosis of ectopic pancreas in the gallbladder, despite its rarity, can be considered.

CONCLUSION

The clinical significance of aberrant pancreatic tissue in the gall bladder needs to be further explored with cooperation between surgeon and histopathologist. Ultrasonography and computed tomography cannot distinguish an aberrant pancreas in the gallbladder from other lesions.

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