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Heterotopic Pancreatic Tissue in Gall Bladder: A Rare Entity

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Abstract

Introduction: Pancreatic heterotopia is a rare pathological entity and its localization in gall bladder is extremely rare. It is mostly asymptomatic incidental finding and rarely gives rise to complications. It can be associated with incidental finding or can be symptomatic or may cause complication. Heterotopic pancreas (HP) also referred to as ectopic pancreas, pancreatic choristoma, or pancreatic rest, is defined as the aberrantly located pancreatic tissue in an anomalous location without any anatomic, vascular, or neural continuity with the main body of the normal pancreas. The most common location for HP is the duodenum, upper jejunum and stomach.

Case report: A 35 year male was admitted to the Surgical Department of Era's Lucknow Medical College and Hospital for an elective laparoscopic cholecystectomy. He had complains of sporadic flatulence after meals and belching. His gall bladder was resected post an USG revealing Gall stones and sent for histopathology where he was diagnosed as a case of heterotopic pancreas in the GB neck with chronic cholecystitis.

Discussion: Localization of pancreatic heterotopia in gall bladder is the second most prevalent pancreatic anomaly but an extremely rare finding. The incidence in gastrointestinal tract is estimated to be from 0.55% to 13.7% on autopsy, and 0.2% in laparatomy. Most commonly these are incidental findings but some studies have implicated them as causative factors in the genesis of cholecystitis, obstructions etc.

Conclusion: HP of the gallbladder may cause clinical symptoms such as cholecystitis as well as complications which can be fatal and hence this condition should be taken into consideration in patients with symptomatic gallbladder disease.

Keywords: Heterotopic pancreas; Cholecystitis; Heterotopia; Haemorrhage

Introduction

Heterotopic pancreas [HP] is a rare pathological entity and its localization in gall bladder is extremely rare. It can be associated with cholecystitis and cholelithiasis Heterotopia is defined as displacement or malpositioning of an organ or a tissue. HP also referred to as ectopic pancreas, pancreatic choristoma, or pancreatic rest, is defined as the presence of pancreatic tissue in an anomalous location without any anatomic, vascular, or neural continuity with the main body of the normal pancreas [1]. HP has been noted in the stomach (24% to 38%), duodenum (9% to 36%), jejunum (0.5% to 27%), ileum (3% to 6%), and Meckel's diverticulum (2% to 6.5%) [2,3]. Despite its congenital origin, pancreatic heterotopia is usually diagnosed during adult life. As it is asymptomatic most of the time, a definitive diagnosis is made on histopathological examination in a gall bladder, removed for other indications [4]. Herein we present a case of HP in gall bladder in a 40-year-old male who presented with clinical findings of cholecystitis [5].

Similar to HP of other organs, HP of the gallbladder itself has no clinical importance and is found incidentally in most cases. However, there have been some reports of pancreas as a cause of gallbladder disease like hemorrhage, obstruction or may be pancreatitis, abscess, cyst or tumors. Therefore in cholecystitis without any pathology, heterotopic pancreas should be taken into consideration before it is diagnosed as idiopathic [6-8].

Case Report

A 35 year male was admitted to the Surgical Department of Era's Lucknow Medical College and Hospital for an elective cholecystectomy. He had complains of sporadic flatulence after meals and belching. On physical examination there was no tenderness or pain, Murphy's sign was negative. His abdominal USG prior to surgery revealed cholelithiasis. His Complete blood count and Liver Function Tests were within normal limits. Patient underwent laparoscopic cholecystectomy. His postoperative period was uneventful and the patient was discharged the next day after the procedure.

Pathological Examination

Grossly, we received an opened up cholecystectomy specimen measuring 5 × 4 × 1 cm. On cut opening the section had a small whitish intramural nodule measuring approximately 1mm at the neck was seen. The rest of the mucosa was greenish velvety with few small blackish stones seen. The wall thickness was 0.3 cm to 0.4 cm. No other gross abnormality seen. On microscopy we saw hyperplastic gall bladder mucosa thrown into folds lined by tall columnar epithelium. The lamina propria was infiltrated with lymphoplasmacytic chronic inflammatory infiltrate and just below the muscularis propria there was presence of aberrant pancreatic tissue consisting of exocrine acini and at the neck area occasional ducts. The remaining sections had features of chronic cholecystitis with mild hyperplasia no dysplasia. No endocrine pancreas was identified. No other granulomatous or neoplastic pathology was identified (Figure 1).

Discussion

Although HP is the second most prevalent pancreatic anomaly, the incidence in gastrointestinal tract is estimated to be from 0.55% to

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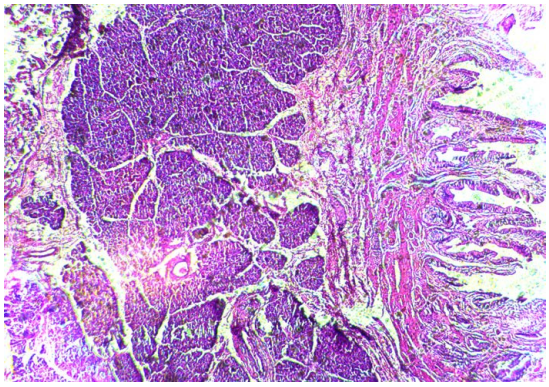

Figure 1: Pathological examination.

13.7% on autopsy, and 0.2% in laparatomy [9,10]. The localization of gall bladder by HP is rare. The majority of cases reported are in stomach (25% to 60%) however other sites are esophagus terminal ileum, colon, meckel's diverticulum, omentum, mesentery, spleen, liver, papilla of vater, bladder, lung, lymphodes [11-13]. Since Poppi's publication in 1916, there have been only 30 more cases of HP worldwide [12,13]. In half of the reported cases, HP is seen to be localized close to the neck of the gallbladder [14]. Similar to this observation, in our patient HP was located in the neck region.

Choristomas are closely related to benign tumors found in abnormal location. While an hamartoma is an excess of normal tissue in normal location choriostoma is excess of abnormal tissue in abnormal locations. Heterotopic pancreas (HP) is defined as the presence of pancreatic tissue that has neither anatomical nor vascular or neural continuity with the main body of the pancreas [15]. HP in the gallbladder is very rarely symptomatic. In most reported cases, it is an incidental pathological findings and coexisting with gallstones [14,15]. However, there have been some reported symptomatic gallbladder diseases due to HP [16] The most commonly occurring complication of HP in gall bladder are hemorrhage and obstruction of the bile duct and gastrointestinal tract [17]. Heterotopic pancreas may be subject to pancreatitis, abscess, or cysts, and pancreatic tumors such as adenocarcinoma, islet cell tumors, it can also be associated with bile duct obstruction, pancreatitis, cysts abscess, calcifications pancreatic intraepithelial neoplasm's and so on [18]. Another case reported by Inceoglu et al. [8] shows a case of HP in the cystic duct with hydrops of the gallbladder and chronic pancreatitis of the ectopic tissue. The preoperative diagnosis of HP in gallbladder is difficult as mostly these cases are asymptomatic or are incidental finding [10,11]. Symptomatology and clinical findings in most cases suggest gallbladder disease, mainly cholelithiasis and cholecystitis [9]. It has been seen that, despite its high resolution, USG is not specific for HP and impossible to distinguish HP from other lesions such as cholesterol polyps, adenoma and carcinoma [17].

The origin of heterotopic pancreatic tissue is controversial but 3 theories have been proposed. One suggests that pancreatic tissue is separated from the main pancreas during embryonic rotation [5,6] the other theory says that during the growth of the ventral pancreatic bud a proportion is transported by the longitudinal growth of the intestines [5,6]. Therefore its presence in the gallbladder might indicate derivation from the ventral diverticulum. The site of organ or tissue formation is also determined by strictly coordinated developmental programs involving interplay between extracellular signaling and intracellular transcriptional factor networks. The Notch and Hes-1 (hairy enhancer of split) are main pathways involved in pancreatic differentiation in developing foregut [6]. Von Heinrich has classified HP into three types microscopically, Type 1: Ectopic tissue with acini, ducts, and islets of Langerhans; Type 2: Ectopic tissue containing only a few acini and ducts, with absent endocrine elements-incomplete arrangement; Type 3: Ectopic tissue with only proliferating excretory ducts and absent exocrine acini and endocrine elements [4].

Malignant transformation of an ectopic pancreas may occasionally occur [16]. However the mechanism by which this aberrant tissue may produce various symptoms is unclear. Previous studies [18-20] have suggested that active pancreatic enzymes, such as amylase and trypsin, refluxing into the biliary tract and gallbladder lumen might produce inflammation, spasm and biliary symptoms in patients without gallstones, and acute cholecystitis in patients with gallstones [16,20-25].

The ductal epithelium of the pancreas is surrounded by smooth muscle component showing reactivity to CK 7, 8, 18, 19 and CA19-9 [Carbohydrate antigen]. No reactivity to CEA, vimentin and CK20 is seen while the pancreatic acini show positivity for alpha1-antitrypsin and alpha1 chymotrypsin. Other endocrinal markers like somatostatin, insulin, chromogranin A are usually negative [16,17].

Conclusion

In conclusion, we can say that, HP in the gallbladder is a very rare condition which is diagnosed incidentally, but may cause clinical symptoms such as cholecystitis as well as complications which can be fatal and hence this condition should be taken into consideration in patients with symptomatic gallbladder disease specially in those without any other specific clinical and laboratory findings before it is diagnosed as idiopathic [6].

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