Case Report

Heterotopic pancreatic tissue in gall bladder incidentally detected in a case of acute cholecystitis: A case report and review of the literature

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ABSTRACT

Heterotopic pancreatic tissue in the gall bladder is a very uncommon lesion. In most cases, it is an incidental finding. The placement of pancreatic tissue in an organ outside the pancreas is called pancreatic heterotopy. Heterotopic pancreas is commonly found in the stomach, small intestine, and Meckel's diverticulum. It has rarely been seen in the gall bladder, bile duct, splenic hilum, or liver. We present a case of incidentally discovered heterotopic pancreatic tissue in a case of acute cholecystitis in a 63-years-old lady having a history of pain in the right hypochondriac region.

Key words: Cholecystitis, Gall bladder, Heterotopic pancreas

Heterotopic pancreatic tissue in the gall bladder is an uncommon lesion. It is the congenital presence of pancreatic tissue outside its normal location in the absence of vascular and anatomical connection with the main pancreas [1]. In most cases, it is an incidental finding. The overall incidence ranges from 0.55% to 13.7% in autopsies [2]. The first reported case of heterotopic pancreas (HP) in gall bladder dates back to 1916 in a description by Otschkin [3]. The gall bladder is a very rare site for HP [1]. About 40 cases have been described worldwide [1].

Here, we present the case of a 63-years-old lady having a history of pain in the right hypochondriac region and were diagnosed with acute calculous cholecystitis.

CASE REPORT

A 63-year-old female presented with a complaint of pain in the abdomen for 3 days. The pain was mainly in the right hypochondriac region, continuous, and dull aching in nature. The patient also complained of nausea and multiple episodes of vomiting. There was no history of fever. The patient was a known case of diabetes mellitus for 10 years and chronic kidney disease for 6 years. The patient had undergone a right renal plasty, 3 months before the current admission.

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On examination, abdominal examination revealed mild tenderness in the right hypochondrium. Serum amylase level was 47 U/L and lipase was 69 U/L. The rest of the routine blood investigations were within normal limits.

Ultrasound imaging showed changes in calculous cholecystitis but no evidence of choledocholithiasis. Magnetic resonance cholangiopancreatography showed tiny calculi and sludge within the gall bladder.

The patient underwent an uneventful laparoscopic cholecystectomy. Intraoperative findings were consistent with cholecystitis (Fig. 1). The patient was discharged on post-operative day 2.

Macroscopic examination revealed a gall bladder measuring 7 × 2 × 1 cm. The external surface was unremarkable. Microscopic examination revealed acute cholecystitis with ectopic pancreatic tissue in the perimuscular adventitia. There was no evidence of malignancy (Fig. 2).

DISCUSSION

HP is the congenital presence of pancreatic tissue outside its normal location in the absence of vascular and anatomical connection with the main pancreas [1]. Different theories have been proposed to explain the origin of HP. The first theory states that HP is formed as a result of pancreatic tissue separation

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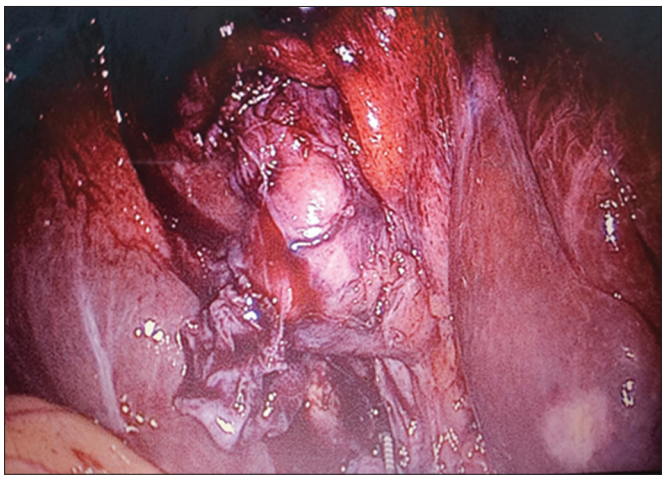

Figure 1: Intraoperative image of Gall bladder showing suspicious region of ectopic pancreatic tissue

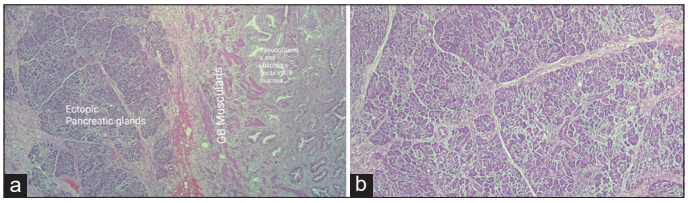

Figure 2: (a) H&E stain showing the presence of pancreatic tissue in gall bladder (×10); (b) H&E stain showing the presence of Type III (exocrine pancreas – acinar tissue only) Fuentes classification (×40)

during embryonic rotation. The second theory suggests that the longitudinal growth of the intestine is responsible for the migration of some cells from pancreatic buds which causes HP in different regions. The third theory which has been advocated recently states that irregularity in the signaling system that decides the destiny of the pancreatic cells, specifically abnormalities in Hairy/enhancer of the split, might be a contributor to the formation of HP in the gall bladder [1].

HP was first described in 1727 by Jean Schultz while von Heinrich provided the first classification of the lesion in 1909 [4]. The classification of ectopic pancreas given by Heinrich *et al*. is as follows [4]: Type I: Mimicking the normal Pancreatic Tissue with the presence of ducts, acini, and endocrine islets. Type II: The presence of a few ducts, a large number of acini, and the absence of endocrine islet. Type III: The presence of a large number of ducts, a few numbers of acini, and the absence of endocrine islets. This classification was modified later by Fuentes in 1973 to include four types of HP [4]. Type I: Resembles the normal pancreatic tissue with the presence of ducts, acini, and endocrine islets. Type II: Canalicular variant with pancreatic ducts only and the absence of acini and endocrine islets. Type III: Exocrine pancreas only with the absence of ducts and endocrine islets. Type IV: Endocrine pancreas only with the absence of canaliculi.

The male to female ratio of HP tissue in the gastrointestinal system is 3:1. However, the literature review shows that the HP in the gall bladder is more common in females due to a higher number of cholecystectomies in females [5]. The presentation of HP in the gall bladder ranges from biliary colic to hydrops gall bladder and even perforation of the gall bladder with peritonitis. The HP tissue is prone to the same pathological conditions as the normal pancreatic tissue such as abscess, pseudocyst formation or chronic pancreatitis, calcification, pancreatic cancer, and endocrine tumors [1,5]. It can cause symptoms related to a location such as jaundice, cholecystitis, and gastrointestinal hemorrhage [5]. Exocrine activity may be associated with acute and chronic cholecystitis and the development of malignant lesions in the biliary tract [6]. Pre-operative radiological examination is not successful in diagnosis. A definitive diagnosis is made after histopathologic examination [6].

Most cases of pancreatic heterotopia have biologically benign behavior; however, only, a few reports have described malignant degeneration and islet cell tumors, cystic tumors, and pancreatic intraepithelial neoplasia can arise from heterotopic pancreatic tissue. Therefore, it is advised to completely excise incidentally discovered pancreatic heterotopia [7].

In our case, the diagnosis was made postoperatively on histopathological examination of the gall bladder. The patient's recovery was uneventful and the patient stays in good health in follow-up care.

CONCLUSION

Heterotopic pancreatic tissue in the gall bladder is a rare pathologic condition that can present as acute cholecystitis. It also has to be considered as a differential in patients labeled as pain due to an unknown cause. Pre-operative diagnosis remains a challenge as of now and it can be diagnosed only on histopathological examination. The significance of the diagnosis of this condition remains unclear due to the limited number of cases being reported in the literature.

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