

CASE REPORT

Heterotopic pancreas of the gallbladder associated with segmental adenomyomatosis of the gallbladder

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Heterotopic pancreas in the gallbladder is extremely rare and usually incidentally discovered at the pathologic examination followed by cholecystectomy for symptomatic gallbladder disease. Up to the presents, only about 30 cases have been reported. We report the case of a 36-year-old female who presented with symptoms of cholecystitis. The histological analysis followed by cholecystectomy revealed heterotopic pancreas of the cystic duct.

Key Words: Heterotopic pancreas, Gallbladder, Cholecystitis

INTRODUCTION

A heterotopic pancreas is defined as the presence of pancreatic tissue outside the boundaries of the pancreas that show no anatomical or vascular connection with the pancreatic structure [1]. The most common site of an ectopic pancreas is the upper gastrointestinal tract [2]. A heterotopic pancreas in the gallbladder is an extremely rare and usually incidentally discovered during a pathologic examination followed by cholecystectomy for symptomatic gallbladder disease. Up to the presents, only about 30 cases have been reported [3,4]. We report a case of heterotopic pancreas which presented with the clinical symptoms of cholecystitis.

CASE REPORT

A 36-year-old female patient visited Pusan National University Hospital complaining of dyspepsia and right upper quadrant discomfort. Physical examination and laboratory results revealed no abnormal findings. The computed tomographic (CT) scan showed a segmental adenomyomatosis (ADM) (Figs. 1, 2). After one year follow-up, the symptoms of the patient had worsened. At that time a laparoscopic cholecystectomy was performed. In the gross finding of resected specimen, the wall of the gallbladder body showed gross focal thickening and luminal narrowing and the cystic duct showed nodular extraluminal thickening. Microscopically the wall of the gallbladder body demonstrated adenomyomatous hyperplasia (Fig. 3). In the examination of the cystic duct normal pancreatic tissue was revealed in subserosal layer, and the pancreatic duct lay on the muscle layer of the cystic duct.

Received August 17, 2012, Revised February 9, 2013, Accepted February 12, 2013

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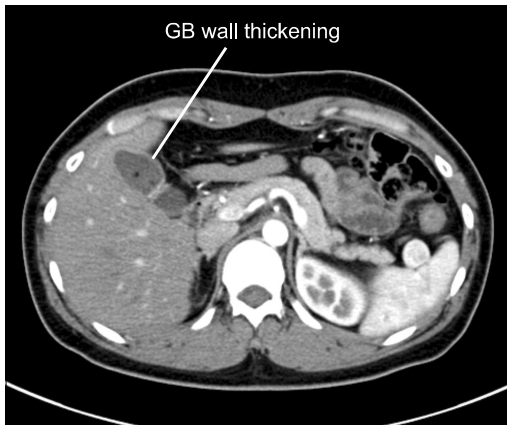


Fig. 1. Preoperative computed tomography findings. Mild gallbladder (GB) wall thickening is observed.

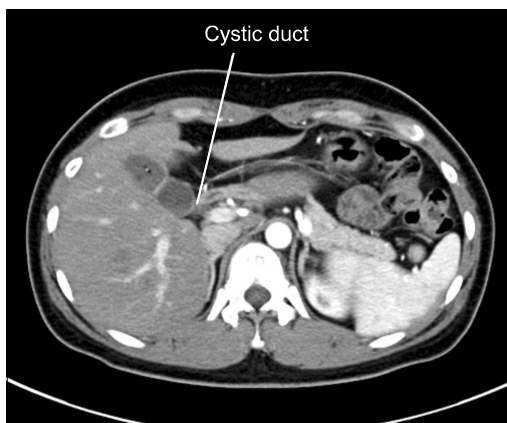


Fig. 2. Preoperative computed tomography findings. Cystic duct abnormalities are not observed.

Also the pancreatic lobules were grouped on the subserosa (Fig. 4). In the high power microscopic findings, the lobules were found to contain acini and islets in normal proportions (Fig. 5).

DISCUSSION

An autopsy series demonstrated a heterotopic pancreas in 0.55% to 13.7% of patients and only 1% had ectopic pancreatic tissue in the gallbladder [5]. Heterotopic pancreas is a congenital anomaly characterized by the growth of aberrant tissue mimicking a normal pancreas without ductal or vascular continuity to the original gland. A recent theory suggested that abnormalities in the Notch sig-

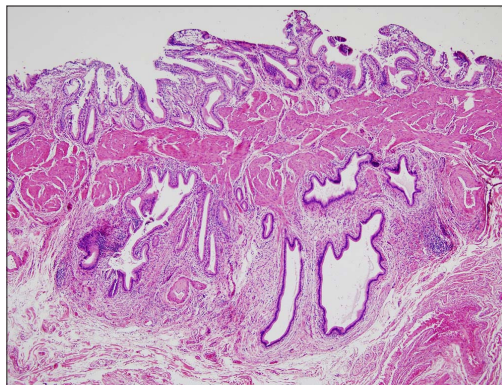


Fig. 3. Adenomatous hyperplasia was present on the body of the gallbladder (H&E, $\times 40$).

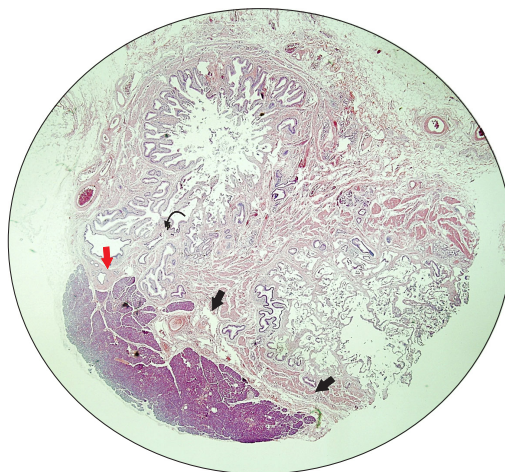


Fig. 4. The pancreatic duct lays on the muscle layer of the cystic duct (red arrow) and the pancreatic lobules are grouped on the subserosa (black arrow) (H&E, $\times 12.5$).

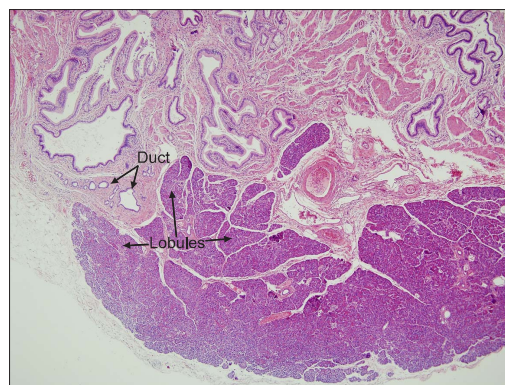


Fig. 5. On high power microscopy, the lobules contain acini and islets in normal proportions (H&E, $\times 40$).

naling system, a main factor for lesion-appropriate pancreatic differentiation in the development of the foregut endoderm, lead to the development of heterotopic pancreatic tissue [6]. But there is no accepted theory that explains the exact origin of a heterotopic pancreas.

A heterotopic pancreas of the gallbladder presents with an exophytic growth that may be similar to polypoid lesions, varying in dimension from a few millimeters to 4 cm. Fifty percent of the lesions arise in the neck of gallbladder and submucosal presentation is the most common form [7]. Histologically, acinar and ductal tissues are always present, whereas islet cells are found in only one third of cases [8]. In our case, the heterotopic pancreas was a 5 mm sized subserosal lesion of the cystic duct and exocrine acinar and ductal components with islets of Langerhans were found on immunohistochemical examinations.

A heterotopic pancreas is asymptomatic and found incidentally after cholecystectomy. Generally, a preoperative diagnosis of heterotopic pancreas in the gallbladder is difficult because of its rarity and nonspecific symptoms. In this case, we could not discriminate whether the patient's symptoms were caused by a segmental ADM or by ectopic pancreas of cystic duct.

During one-year-follow up of segmental ADM, gallbladder wall thickness had not changed but the patient's symptoms had worsened. All symptoms of the patient had disappeared after a cholecystectomy. A high level of pancreatic enzyme in gallbladder bile with an ectopic pancreas can cause pain and eventually damage the gallbladder mucosa, leading to the development of gallbladder cancer [9,10]. Therefore an ectopic pancreas of the gallbladder may be a risk factor of the development of gallbladder cancer.

In the patients who show the symptoms of benign gallbladder disease without a definitive associated lesion in imaging studies, the consideration of possible presence of

ectopic pancreas of gallbladder is recommended for the diagnosis of the disease, even though it is very rare instance.

CONFLICTS OF INTEREST

No potential conflict of interest relevant to this article was reported.

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