INTRODUCTION

Heterotopic pancreas (HP) is defined as the presence of pancreatic tissue lying outside its normal location and lacking anatomical or vascular continuity with the pancreas proper.[1] In 85–90% of reported cases, HP has been found in stomach, duodenum, upper jejunum, whereas its presence in the gallbladder is very rare.[1,2] Despite its congenital origin, pancreatic heterotopia is usually diagnosed during adult life.[3,4] As it is asymptomatic most of the time, a definitive diagnosis is made on histopathological examination in a gall bladder, removed for other indications.[1,5] We report a case of HP of the gallbladder along with pseudopyloric metaplasia and segmental adenomyomatous hyperplasia in a 45-year-old woman. Up to the present study, about 31 cases of HP in the gallbladder have been reported,[5] but this is the first of its kind which has two other histopathological findings associated with it.

CASE REPORT

A 45-year-old woman presented to our hospital with 2 months history of the right upper quadrant abdominal pain along with nausea and vomiting. Her history was unremarkable. All her vitals were stable. Routine blood investigations including renal and liver function tests revealed no abnormality. On physical examination, there was tenderness in the right upper quadrant. Abdominal ultrasound showed no abnormality. Based on the diagnosis of cholecystitis, cholecystectomy was carried out.

On gross examination, the gall bladder measured 7 cm in length and 2.5 cm in circumference with a wall thickness ranging from 0.2 to 0.4 cm. The serosa was unremarkable. Cut section revealed velvety green mucosa.

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On microscopic examination, there was a well-circumscribed rest of heterotopic pancreatic tissue on the serosal aspect of the gall bladder wall, composed of lobules of exocrine pancreatic acini and an occasional duct. Islets of Langerhans were not seen. Mucosal layer adjacent to the pancreatic tissue showed adenomyomatous hyperplasia [Figure 1]. A focus elsewhere showed pseudopyloric metaplasia apart from histological features of chronic cholecystitis [Figure 2].

The diagnosis was thereby established as chronic cholecystitis with heterotopic pancreatic tissue, pseudopyloric metaplasia, and segmental adenomyomatous hyperplasia of the gall bladder.

The patient’s postoperative course was uneventful and was discharged after 5 days without any complications. After a follow-up of 2 months, the patient was asymptomatic and had recovered fully.

**DISCUSSION**

Although HP is the second most prevalent pancreatic anomaly, the incidence in gastrointestinal tract is estimated to be from 0.55% to 13.7% on autopsy and 0.2% on laparotomy.\(^5\) Despite the frequent occurrence of HP in the stomach, duodenum, and upper jejunum, the gallbladder localization is extremely rare.\(^7\)

Heterotopic tissue is usually located in the neck or fundus of the gall bladder, varies in size from 0.1 to 1.0 cm, and may exhibit several patterns, ranging from intramural to exophytic to polypoidal lesions.\(^3\)\(^6\) As there is no submucosal layer in the gall bladder, HP is usually seen in the muscularis. Microscopic examination shows a varying degree of excretory ducts, exocrine glands, and islets of Langerhans.\(^8\) Microscopically, HP has been classified into three types by von Heinrich - Type 1: Ectopic tissue with acini, ducts, and islets of Langerhans; Type 2: Ectopic tissue containing only a few acini and ducts, with absence of endocrine elements - incomplete arrangement; Type 3: Ectopic tissue with only proliferating excretory ducts and absence of exocrine acini and endocrine elements.\(^9\) Our case was considered to be Type 2, based on the Heinrich classification.

A recent theory suggested that abnormalities in the notch signaling system, a main factor for lesion-appropriate pancreatic differentiation in the development of the foregut endoderm, lead to the development of heterotopic pancreatic tissue. However, there is no accepted theory that explains the exact origin of an HP.\(^5\)

**CONCLUSION**

Chronic cholecystitis with heterotopic pancreatic tissue, pseudopyloric metaplasia, and adenomyomatous hyperplasia of the gall bladder is rarely encountered. HP of the gall bladder itself is a very rare condition which is usually diagnosed incidentally, but may cause symptoms of benign gall bladder disease without a definitive lesion on radiology. Awareness of this under-reported condition may help in its recognition and this, in turn, may shed more light on its clinical significance. In our case, we could not discriminate whether the patient’s symptoms were caused by HP or by segmental adenomyomatous hyperplasia of the gall bladder.

![Figure 1: Photomicrograph of gall bladder showing a well-circumscribed rest of heterotopic pancreas (arrowhead) and adjacent mucosa showing adenomyomatous hyperplasia (arrow) (H and E, ×40). Inset shows lobules of pancreatic acini and an occasional duct (H and E, ×400)](image1)

![Figure 2: Photomicrograph showing pseudopyloric metaplasia in a separate focus along with features of chronic cholecystitis (H and E, ×40)](image2)
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Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Armstrong CP, King PM, Dixon JM, Macleod IB. The clinical significance of heterotopic pancreas in the gastrointestinal tract. Br J Surg 1981;68:384-7.
2. Dolan RV, ReMine WH, Dockerty MB. The fate of heterotopic pancreatic tissue. A study of 212 cases. Arch Surg 1974;109:762-5.
3. Elhence P, Bansal R, Agrawal N. Heterotopic pancreas in gall bladder associated with chronic cholecystolithiasis. Int J Appl Basic Med Res 2012;2:142-3.
4. Guer H, Bagci P, Coskunoglu EZ, Karadag C. Heterotopic pancreatic tissue located in the gallbladder wall. A case report. JOP 2011;12:152-4.
5. Lee SW, Yun SP, Seo HJ. Heterotopic pancreas of the gallbladder associated with segmental adenomyomatosis of the gallbladder. J Korean Surg Soc 2013;84:309-11.
6. Inceoglu R, Doshuglu HH, Kullu S, Ahiskali R, Doslu FA. An unusual cause of hydropic gallbladder and biliary colic – Heterotopic pancreatic tissue in the cystic duct: Report of a case and review of the literature. Surg Today 1993;23:532-4.
7. Elpek GO, Bozova S, Küpesiz GY, Ogüs M. An unusual cause of cholecystitis: Heterotopic pancreatic tissue in the gallbladder. World J Gastroenterol 2007;13:313-5.
8. Pang LC. Pancreatic heterotopia: A reappraisal and clinicopathologic analysis of 32 cases. South Med J 1988;81:1264-75.
9. Ogata M, Chihara N, Matsunobu T, Koizumi M, Yoshino M, Shioya T, et al. Case of intra-abdominal endocrine tumor possibly arising from an ectopic pancreas. J Nippon Med Sch 2007;74:168-72.