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Case Report

The rare entity of ectopic pancreatic tissue in the gallbladder: A case report

Georgios Chatziantoniou [a,1], Georgios Tzikos [a,1,*], Aristeidis Ioannidis [a], Antonia Loukousia [b], Georgia Raptou [b], Antonios Michalopoulos [a], Daniel Paramythiotis [a]

[a] 1st Propaedeutic Department of Surgery, AHEPA University Hospital, Aristotle University of Thessaloniki, St. Kiriakidi 1, 54621, Thessaloniki, Greece
[b] Pathology Department, Faculty of Medicine, Aristotle University of Thessaloniki, Thessaloniki, Greece

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ABSTRACT

Introduction: and importance: Ectopic pancreatic tissue (EPT) is a rare clinical entity, which is defined as the presence of pancreatic tissue without any anatomic or vascular connection with the main body of the pancreas. EPT could be found anywhere in the gastrointestinal tract; most commonly in stomach. The aim of this study is to present a rare case report of EPT located in the gallbladder.

Case presentation: A 37-year-old woman was scheduled in our surgical department for elective laparoscopic cholecystectomy due to symptomatic chololithiasis. Preoperative ultrasound imaging was indicative only for the presence of multiple stone tin the gallbladder's fundus. The patient had an uneventful recovery and discharged the first postoperative day. Surprisingly, the final pathology report of the specimen referred the existence of EPT in the subserosa territory, as an incidental finding.

Clinical discussion: EPT is almost impossible to be diagnosed preoperatively due to its various clinical presentation and the low discriminating power of all the usual imaging tests. However, given the potential malignant transformation of the EPT, physicians should be aware of this clinical entity and consider cholecystectomy immediately when it is highly suspected.

Conclusion: EPT in the gallbladder is a very rare finding. There are no established "gold standard" techniques to identify it preoperatively. The patients are either asymptomatic or presenting with non-specific symptoms and almost always the pathology examination, after cholecystectomy, establishes the definite diagnosis.

1. Introduction

Ectopic pancreatic tissue or pancreatic heterotopia (EPT) is an uncommon congenital abnormality characterized by the presence of pancreatic tissue outside of the anatomical site of the pancreas. The term derives from the combination of the two Greek words "hetero-" which means "other" and "-topia" which means "site", noting the abnormal location of pancreatic cells. EPT can be detected anywhere along the gastrointestinal tract (GI), primarily in the stomach, the duodenum and the colon [1]. However, it can also be found in less common sites such as the spleen, the liver, the omentum, the lungs, the umbilicus, the fallopian tube, the tongue, the esophagus, and the gallbladder [2]. EPT incidence is estimated to be approximately one per 500 surgical operations involving the upper GI tract [1]. In this study, we present a rare case report of a patient having undergone elective laparoscopic cholecystectomy and the histopathological examination of the specimen unveiled EPT in the gallbladder, as an incidental finding. This case report has been reported in line with the SCARE Criteria [3].

2. Presentation of case

A 37-year-old woman referred to our outpatient department for surgical evaluation due to chronic pain in the right upper quadrant of the abdomen, deteriorating especially after fatty meals. Her medical history was clear without taking any medication, while she was a smoker (about 10ciggaretes/day). No allergies were reported. During the clinical examination, her vital signs were within normal ranges (Blood pressure:130/85 mmHg; Heart rate: 88 pulses/min; Respiration rate: 13 breaths/min; Temperature: 36.6 °C; Oxygen Saturation: 98%). However, the palpation of the abdomen revealed tenderness in the right upper quadrant with a negative Murphy sign. The patient brought us her recent laboratory evaluation which was normal, except the elevated levels of

* Corresponding author. 4 Souliou Str, Kalamaria, Thessaloniki, Greece.
E-mail address: tzikos_giorgos@outlook.com (G. Tzikos).
[1] These authors have contributed equally to this work and share first authorship

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Gamma-Glutamyl Transpeptidase (γ-GT: 242 U/I, reference range: 5–40 U/I). Additionally, she had already undergone ultrasonographic evaluation of the upper abdomen reporting a gallbladder with thin wall, containing multiple stones in the fundus (Fig. 1). As a result, the patient was scheduled for laparoscopic cholecystectomy, under the diagnosis of symptomatic chololithiasis [4]. The operation was held two weeks later by the hepatobiliary specialist surgeon of our department with the help of two junior trainees with about 3 years of surgical specialty training. The patient had an uncomplicated postoperative period and discharged the next day of the operation with order to follow a free fat diet for one month. In addition, the γ-GT levels were normalized five weeks postoperatively and, 6 months postoperatively, the patient did not report any delayed complications or symptoms, as it was expected.

During pathology examination, macroscopic examination revealed a gallbladder measuring 7.6 × 2.8cm, containing multiple small stones. The average wall thickness was 0.7cm. The mucosa showed yellow flecks against green background (strawberry gallbladder). In the neck region, a single solid, yellowish, intramural nodule measuring 1.2cm was identified. Multiple representative sections were analyzed. Microscopic examination showed characteristics indicative of chronic cholecystitis with cholesterolosis. Regarding the nodule described macroscopically, histology confirmed the presence of heterotopic pancreatic tissue beneath the muscularis propria (Fig. 2A). Both exocrine and endocrine pancreatic tissues were present, composed of acini and ducts along with a few islets of Langerhans (Fig. 2B). No evidence of dysplasia or malignancy was found in any of the sections examined. All these findings confirmed EPT in the gallbladder, as an incidental finding. In view of the fact that no malignancy had been reported after the pathology examination and the amelioration of the patient's clinical condition described above, there was no need for further actions regarding her treatment.

3. Discussion

In this study we presented a case of EPT in the gallbladder, which was documented only after cholecystectomy performed due to symptomatic chololithiasis. In general, EPT is a rare finding. The most common sites of EPT are the stomach and the duodenum [1]. The etiology of this abnormality has not been defined yet. Nevertheless, it is believed that EPT derives from the early separation of the pancreas during the rotation of the gastrointestinal tract in the embryonic period [5]. Another theory supports that the suppression of the transcription factor HES-1 (Hairy and enhancer of split 1) results in abnormal signaling of the developing foregut endoderm tissue during embryogenesis and contributes to EPT in the gallbladder [6,7].

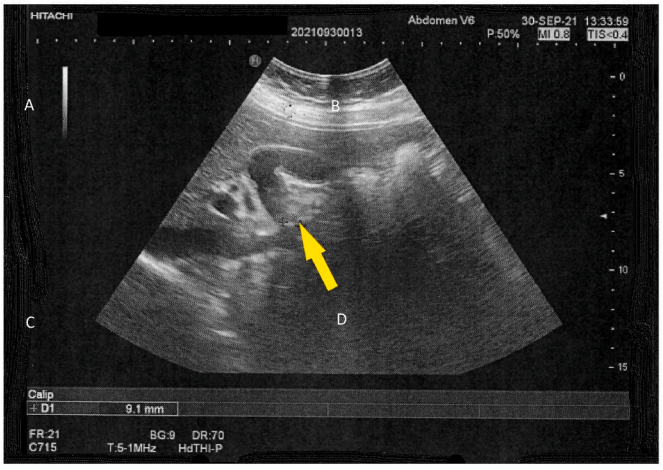

Fig. 1. Ultrasound image showing the gallbladder containing multiple stones (yellow arrow) in the fundus. (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

Although, the male-to-female ratio of any type of EPT is 3:1, specifically for EPT in the gallbladder region, there is a female predominance, probably due to more cholecystectomies among women [8,9]. The median age of the reported cases was 46, with most patients being more than 40, contrary to our case, while chololithiasis coexisted with EPT in half of them. Less than 40 cases of EPT in gallbladder have already been reported in the literature [9]. In almost all the cases, EPT in the gallbladder is diagnosed, after the resection and the histopathologic examination of the gallbladder. This clinical entity most frequently causes no symptoms. However, it could imitate other medical conditions such as chololithiasis, acute or chronic cholecystitis, gallbladder polyps-hydrops, adenomyoma, or carcinoma. In addition, gallbladder perforation with peritonitis, due to EPT ulceration, has also been reported [10]. Moreover, EPT could induce the same pathological events as the normal pancreatic tissue which includes cysts, pseudocysts formation, abscess and acute or chronic pancreatitis. Furthermore, it is considered that pancreatic enzymes secreted from EPT could affect and degenerate gallbladder mucosa potentially leading to gallbladder cancer [11,12]. Therefore, in rare cases which EPT in the gallbladder could be diagnosed preoperatively, cholecystectomy should be performed as the definite treatment preventing of any potential malignant transformation. Our patient had symptoms resembled to chronic cholecystitis and, based on the laboratory and imaging findings, EPT was not included in our differential diagnosis.

Regarding the preoperative imaging diagnosis of EPT, the ultrasound or the computed tomography (CT), have low discriminatory power without contributing efficiently in the recognition of this entity, because both of them are not able to distinguish EPT from polyps, stones or carcinoma [13]. In our case, the preoperative imaging assessment with ultrasound did not reveal EPT, too. Thus, the definite diagnosis was established by the pathologist, after laparoscopic cholecystectomy had been performed. Macroscopically, EPT in the gallbladder may present as an exophytic mass, similar to polypoid lesions, or as nodule with yellow-colored appearance and its size varies from a few millimeters to even 4cm [14].

In 1909, Heine Von Heinrich initially proposed a classification system into 3 groups, based on the pathological findings of EPT, which was then modified by Gaspar Fuentes et al., in 1973 (Table 1) [15,16]. The classification of Gaspar Fuentes et al. includes 4 types of EPT and given that in our case the pathology report revealed the presence of acini, ducts and islet-like pancreatic gland, it was classified as type I EPT in the gallbladder, following Fuentes' classification.

The strength of this case report is that it is another reported case of EPT in the gallbladder which confirms the inconsistency between the preoperative imaging tests and the final diagnosis of EPT in the gallbladder, a fact which all the clinicians should always have in mind. On the other hand, the limitation of the study is that our patient presented with the classical presentation of symptomatic chololithiasis, without any novel clinical symptom or any other disorder, which could make this case unique. However, even this case adds some extra information about the under-reported entity of EPT in the gallbladder.

4. Conclusion

EPT in the gallbladder remains a rare clinical entity. Until now, there are no settled clinical or imaging techniques for diagnosing this under-reported medical condition and only awareness and further research may enlighten more its clinical significance.

Ethical approval

Our case report obtained ethics approval from the ethics committee of our hospital and the patient gave her informed consent to participate.

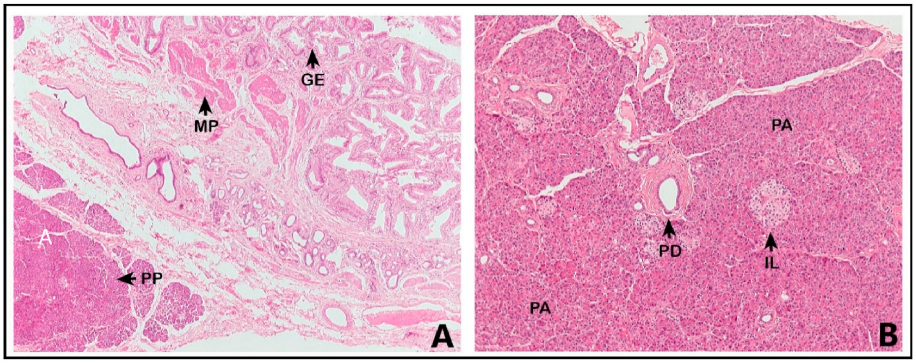

Fig. 2. A: Heterotopic pancreatic tissue in gallbladder (hematoxylin and eosin stain, 4X) showing: surface epithelium of gallbladder (GE) with the pancreatic parenchyma (PP) located beneath the muscularis propria (MP); B: Pancreatic heterotopia Type I in gallbladder (hematoxylin and eosin stain, 10X) showing: pancreatic acini (PA), pancreatic duct (PD), and islet of Langerhans (IL).

Table 1
Classification system for pancreatic heterotopia by Gaspar Fuentes et al.

Type I	Presence of acini, ducts and islet-like pancreatic gland
Type II	Canalicular variant with pancreatic ducts
Type III	Exocrine pancreas with acinar tissue
Type IV	Endocrine pancreas cellular islets

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Author contribution

GC and GT wrote the manuscript and collected the data. AL and GR collected also the data for the study and performed the pathology analysis. AI revised the manuscript for grammar and syntax mistakes. DP corrected the manuscript for its scientific basis. AM was the director of the Department of Surgery and the consultant surgeon who provided the case. All authors have read and approved the final manuscript.

Research registration

None declared.

Guarantor

DP accepts full responsibility for the work and the conduct of the study, had access to the data, and controlled the decision to publish.

Availability of data and materials

The data and materials/figures used in the current study are available from the corresponding author on reasonable request.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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