

Ectopic liver tissue on gallbladder (choristoma): Two case reports

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ABSTRACT

Ectopic liver tissue is a rare developmental anomaly and is most commonly seen on the gallbladder. A 45-year-old male and 41-year-old female patient underwent laparoscopic cholecystectomy due to gallstones, and ectopic liver tissue was found on the patients' gallbladder. During the operation, ectopic liver tissue removed with the surgical specimen material. Pathology report is compatible with the usual liver parenchyma. Discussion: Although there is no strong information about the long-term follow-up results of the ectopic liver, surgical resection is recommended due to the high probability of carcinogenesis (hepatocellular carcinoma). However, this possibility is lower in localizations on the gallbladder. Therefore, if there is a pathology in the gallbladder, its excision with the gallbladder is recommended.

Keywords: Ectopic liver tissue; gallbladder; hepatic choristoma; hepatocellular carcinoma; liver.

Introduction

Ectopic liver tissue, a developmental anomaly that is rarely encountered in the literature, also called heterotopic liver or choristoma.^[1] Although it can be seen on many organs due to embryological development, it is most commonly observed on the gallbladder; less frequently in the umbilical cord, hepatic ligament, stomach, retroperitoneum, and thorax.^[2]

Ectopic liver tissue is mostly asymptomatic. It is encountered incidentally during the operation or at autopsy. It can cause complications such as torsion, malignant transformation, peripheral organ compression, and intraperitoneal bleeding.^[3] Although the histological structure of ectopic liver tissue is similar to that of normal liver, it has

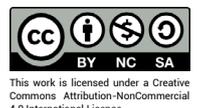
no metabolic function and is prone to carcinogenesis.^[4] In this article, we presented two cases of ectopic liver tissue, one in the gallbladder serosa and the other in the gallbladder mesentery.

Case Report

Case 1 – Gallbladder stones were detected on ultrasound in a 45-year-old male patient who had complained of biliary colic for the past 1 year and elective laparoscopic cholecystectomy was performed. During the operation, a smooth brown tissue with a size of 16 mm × 8 mm × 3 mm attached to the gallbladder serosa with a thin meso-tissue was detected (Fig. 1). Tissue was removed together with the gallbladder (Figs. 2, 3). Histological examina-



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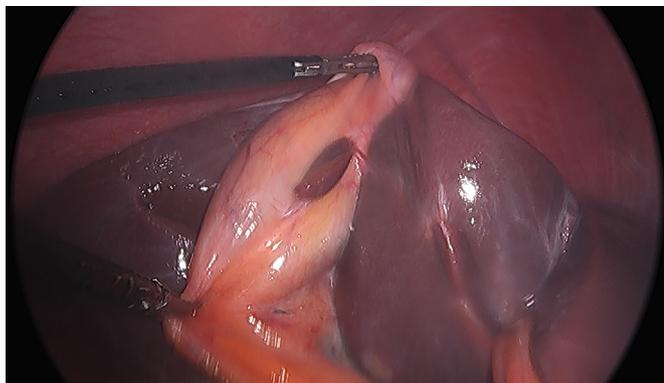


Figure 1. During laparoscopic cholecystectomy, a brown tissue attached to the gallbladder serosa with a thin meso-tissue was detected.



Figure 2. After it was found out that it was an ectopic liver tissue, it was removed laparoscopically with the specimen.



Figure 3. Gallbladder and heterotopic tissue specimens.

tion revealed normal liver tissue containing normal tissue elements (Fig. 4). The patient, who had an uneventful postoperative recovery period, was discharged after 48 h.

Case 1 – A 41-year-old female patient, who had a history of biliary colic for 6 months, underwent endoscopic retrograde cholangiopancreatography on detection of stones in the common bile duct. After the procedure, laparoscopic cholecystectomy was performed due to gallstones. During the operation, double cystic arteries were detected in the vascular structure of the gallbladder, apart from that, an ectopic liver tissue of 8 mm × 4 mm × 2 mm was found attached to the gallbladder wall and was removed together with the gallbladder. Histology report revealed normal liver tissue containing normal hepatocytes (Figs. 5, 6).

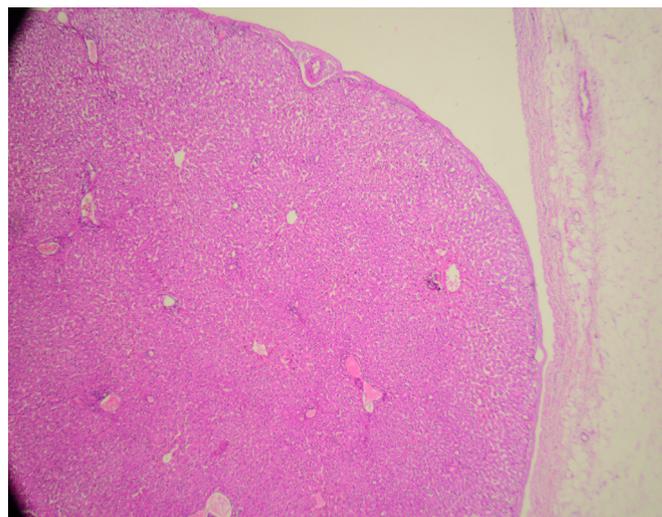


Figure 4. Encapsulated ectopic liver tissue consisting of central vein and surrounding hepatocytes, not showing connection with the liver.

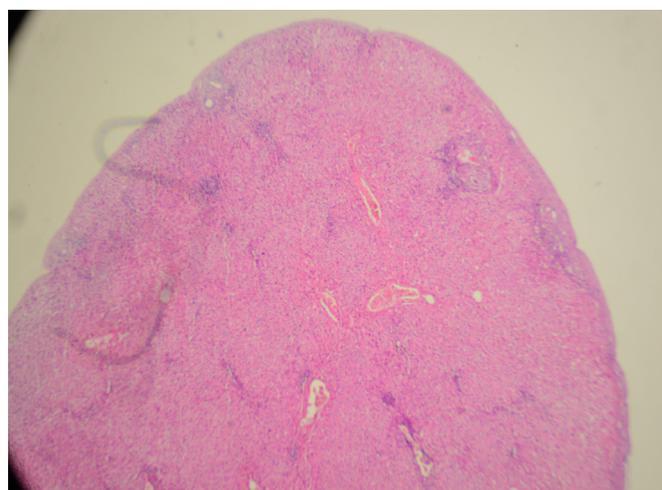


Figure 5. Ectopic tissue showing normal liver morphology.

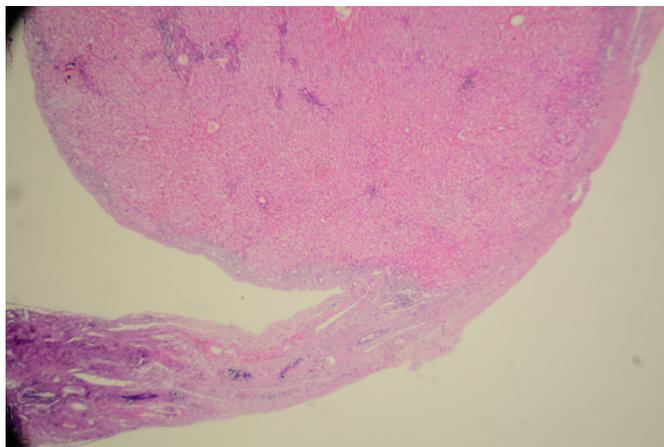


Figure 6. Ectopic liver tissue with its own pedicle and capsule.

Discussion

Heterotopia is the abnormal localization of a well-differentiated normal tissue. Heterotopic tissue can be seen throughout the entire intestinal tract from mouth to rectum. In the literature, the presence of heterotopic gastric mucosa, intestinal mucosa, pancreatic, and liver tissue in the gallbladder has been reported.^[5,6]

The incidence of ectopic liver tissue attached to the gallbladder wall is unknown, as it is usually asymptomatic; most cases are detected during laparotomy, laparoscopy, and autopsy.^[7] In the literature, the incidence is defined as 0.24–0.47%.^[8]

There are several theories explaining the formation of ectopic liver tissue. These theories include atrophy of the connection of an accessory liver lobe with the main liver during the embryological period, or regression of this connection, migration, or misplacement of a part of the pars hepatica in the liver bud, dorsal budding of the liver tissue before the closure of the pleuroperitoneal canal, the presence of mesenchymal cells transforming into hepatocytes in a different location than they should be, and cell clusters remain in the foregut region after the diaphragm or umbilical ring closes.^[9]

When heterotopic tissue (ectopic liver) is not found in the gallbladder, it can sometimes be seen with other congenital anomalies; such as biliary atresia, caudate lobe agenesis, omphalocele, biliary tract cysts or heart, and truncal anomalies.^[10] In the second case, ectopic liver tissue was found to be accompanied by double cystic arteries, and it is thought to be caused by developmental changes in the primitive ventral splanchnic arteries.^[11] This suggests that it is due to the ability of the multipotent cells of the endoderm and mesoderm to change to form different layers.

^[12] However, in our case reports, the congenital anomalies mentioned were not encountered.

Ectopic liver tissue may have a mesentery with its own vessels. Depending on its location, it may drain into the biliary system or other organs. In most cases, it does not have a drainage system. Arterial blood supply of ectopic liver tissue is often provided by an autonomic artery that does not originate from the hepatic artery. In addition, it does not have its own portal vein system and ductal system connected to the biliary tree.^[13]

Ectopic liver tissue is often asymptomatic, and pre-operative detection is very rare.^[14] Nevertheless, there are cases in the literature that have reported recurrent abdominal pain, intraperitoneal bleeding, hemorrhagic necrosis, surrounding organ compression, esophageal obstruction, portal vein obstruction, and neonatal gastric outlet obstruction due to torsion of ectopic liver tissue.^[15,16] In addition, cirrhosis, fattening, hemosiderosis, and metastatic tumors can be observed, as in normal liver.^[4]

Since ectopic liver tissue is small and rare, it is difficult to detect by preoperative diagnostic methods. It is currently impossible to preoperatively distinguish ectopic liver tissue from other gallbladder lesions such as cholesterol polyps, adenoma, and carcinoma in gallbladder masses. However, ectopic liver tissue should be considered in focal thickening in the gallbladder wall with the same features as the liver in abdominal ultrasonography or CT. With ultrasound-guided biopsy, a definitive diagnosis can be made histopathologically in ectopic liver tissue.^[17]

It is suggested that ectopic liver tissue increases the risk of developing cancer. Hepatocellular carcinoma developed in 22 of 48 cases (located outside the gallbladder), but cancer developed in only one of the 33 cases of ectopic liver tissue attached to the gallbladder. The most likely explanation for this difference is ectopic liver tissue attached to the gallbladder is well differentiated because it occurs at late embryogenesis.^[18]

Conclusion

Ectopic liver tissue is a rare developmental anomaly. It's mostly detected incidentally during the laparoscopic gallbladder operations, and pre-operative diagnosis is difficult. Although it is known that ectopic liver tissue is usually asymptomatic, it has risks such as risk of malignant transformation (higher risk outside of the gallbladder), bleeding, and torsion; the inclusion of the ectopic tissue

in the surgical specimen is considered to be the most appropriate procedure during the operation.

Disclosures

Informed Consent: Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

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References

1. Akbulut S, Demyati K, Ciftci F, Koc C, Tuncer A, Sahin E, et al. Ectopic liver tissue (choristoma) on the gallbladder: A comprehensive literature review. *World J Gastrointest Surg* 2020;12:534–48. [\[CrossRef\]](#)
2. Griniatsos J, Riaz AA, Isla AM. Two cases of ectopic liver attached to the gallbladder wall. *HPB (Oxford)* 2002;4:191–4.
3. Kachi A, Bou Rached C, El-Helou E, Kanj M, Kansoun AH. Incidental finding of ectopic liver during laparoscopic cholecystectomy. *Am J Case Rep* 2020;21:e921410. [\[CrossRef\]](#)
4. Martinez CA, de Resende HC Jr, Rodrigues MR, Sato DT, Brunialti CV, Palma RT. Gallbladder associated ectopic liver: A rare finding during a laparoscopic cholecystectomy. *Int J Surg Case Rep* 2013;4:312–5. [\[CrossRef\]](#)
5. Akay T. Foregut-origin ciliated cyst of the gallbladder. *Turkiye Klinikleri J Case Rep* 2019;27:9–12. [\[CrossRef\]](#)
6. Hamazaki K, Fujiwara T. Heterotopic gastric mucosa in the gallbladder. *J Gastroenterol* 2000;35:376–81. [\[CrossRef\]](#)
7. Weber-Alvarez P, Weber-Sánchez LA, Carbó-Romano R, Garteiz-Martínez D. Laparoscopic treatment of hepatic choristoma in the gallbladder wall: A clinical case presentation and literature review. *Rev Gastroenterol Mex* 2017;82:189–90. [\[CrossRef\]](#)
8. Arslan Y, Altintoprak F, Serin KR, Kivilcim T, Yalkin O, Ozkan OV, et al. Rare entity: Ectopic liver tissue in the wall of the gallbladder-A case report. *World J Clin Cases* 2014;2:924–6.
9. Aslan F, Akkas G. Ectopic liver attached to the gallbladder wall: a rare finding during laparoscopic cholecystectomy. *Annals of Med Medical Educ* 2016;3:23–7.
10. Park WH, Choi SO, Lee SS, Randolph JG. Ectopic umbilical liver in conjunction with biliary atresia: Uncommon association. *J Pediatr Surg* 1991;26:219–22. [\[CrossRef\]](#)
11. Akay T, Leblebici M. Incidence of double cystic artery: a clinical study. *Int Surg J* 2020;7:2837–42. [\[CrossRef\]](#)
12. Koh CE, Hunt R. Ectopic liver encountered during laparoscopic cholecystectomy. *Asian J Surg* 2007;30:227–9.
13. Leone N, Saettone S, De Paolis O, Carucci P, Brunello F, De Angelis C, et al. Ectopic livers and related pathology: report of three cases of benign lesions. *Dig Dis Sci* 2005;50:1818–22.
14. Granek RJ, Battye S, Wijeratne S. Ectopic liver attached to a chronically inflamed gallbladder: a rare and surgically challenging combination. *ANZ J Surg* 2019;89:388–9. [\[CrossRef\]](#)
15. Corbitt N, Eric JR, Marta HS, Dai HC. Accessory hepatic lobes in the pediatric population: A report of three cases of torsion and literature review. *JPS Case Reports* 2017;16:15–8.
16. Zhang ZL, Wang JL, Guo CL, Li Q, Li L, Pang Y, et al. Ectopic liver tissue in the esophagus: A case report. *Medicine (Baltimore)* 2019;98:e15260. [\[CrossRef\]](#)
17. Hamdani SD, Baron RL. Ectopic liver simulating a mass in the gallbladder wall: Imaging findings. *ARJ* 1994;162:647–8.
18. Arakawa M, Kimura Y, Sakata K, Kubo Y, Fukushima T, Okuda K. Propensity of ectopic liver to hepatocarcinogenesis: case reports and a review of the literature. *Hepatology* 1999;29:57–61. [\[CrossRef\]](#)