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Invagination of Transformed Confluence Cysts of Lobar Ducts into the Common Hepatic Duct

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Abstract: *Rationale*: Cystic transformation of the bile ducts is rare. There are no publications on the complication of this pathology in the form of invagination of hepatic duct cysts. The search for optimal methods of surgical treatment of transformed cysts continues. *Objective*: To analyze the surgical treatment of patients with bile duct cysts, their complications and the choice of optimal methods of surgical correction depending on the localization of cysts. *Material and methods*: The 40–year experience of treating 15 women and 1 man with cystic transformation of the bile ducts is presented. There were 2 rare observations – the invagination of fragments of partially excised walls of confluence cysts of lobar ducts through their iatrogenic defect into the common hepatic duct. The main diagnostic methods were ultrasound, endoscopic retrograde cholangiopancreatography, magnetic resonance cholangiopancreatography. *Results*: For intrahepatic cysts, left-sided hemihepatectomy (1), resection of 2 and 3 segments of the liver with simultaneous excision of all walls of the hepaticocholedoch cyst, hepaticoejunoanastomosis with isolated jejunum according to Roux (1) was performed. Invagination of fragments of partially excised walls of cysts confluence of lobar ducts through their iatrogenic defect into the common hepatic duct was eliminated using disinvagination through hepaticotomy (1) and resection of hepatic ducts with invaginate with the creation of a bigepaticoejunoanastomosis into the intestinal ring of the isolated jejunum (1). The walls of the cysts of the ducts of the subhepatic localization were completely excised (11) using bigepaticoejunoanastomosis into the intestinal ring of the isolated jejunum (2), hepaticoejunostomy according to Roux (9). Cystoenteroanastomosis according to Brown was performed in 2 patients. Regarding the membranous intra-current septa, their excision (1) and resection of the stenosed left hepaticoejunoanastmosis (1) were required. There were no fatal outcomes. Unresectable tumors of the subhepatic space were found in 2 patients 13 and 15 years after complex excision of cysts with hepaticoejunostomy. *Conclusion*: We use active surgical tactics: complete excision of the cyst walls, the creation of a hepaticoejunoanastomosis using an isolated intestinal loop according to Roux and a resection technique for single-lobar liver lesion with cysts. Diagnosis of invagination of cystic fragments into the ducts requires highly informative equipment, treatment requires specialized surgical care.

Keywords: Bile Ducts, Cyst, Invagination, Surgery

1. Introduction

Cystic transformation of the bile ducts is a rare and complex anomaly of the development of the bile ducts. It occurs in the large bile ducts of all parts of the biliary tract, more than 80% is observed in the common hepatic and common bile duct (hepaticocholedoch) [1]. The term transformation is of Latin origin: transformation, change of genetic properties. If we take into account the similarity of hepatic and subhepatic cysts according to clinical and morphological indicators, their heredity by autosomal recessive trait [2, 3] and high numbers of detection of cysts in childhood (60%) [4], then the term

cystic transformation is more pathognomonic for bile ducts cysts. There is no consensus on the causes of cystic transformation, there is no uniform terminology and classification [3, 5, 6]. There are high numbers of strictures of cystodigestive anastomoses and malignancy of cysts [7, 8]. An approach to the formation of various biliodigestive anastomoses is discussed [1, 5, 6, 9, 10].

Cysts of the bile ducts often manifest symptoms of cholelithiasis and its complications: pain in the liver, fever, chills, jaundice, itching. Patients may have gallstones, cholangitis, strictures of the anastomoses. Intrahepatic localization of cysts was isolated in J. Caroli's disease (1958) [11], however, there is a significant commonality between hepatic and subhepatic localization. 1. The congenital nature of cysts that are inherited by an autosomal recessive trait [2, 3]. 2. Large bile ducts are affected. 3. The possibility of combining intrahepatic and subhepatic localizations of cysts [12]. 4. Similarity of clinical symptoms. 5. Similarity of morphological features. 6. High degree of malignancy [7, 8]. Based on the literature data, it can be assumed that the transformation of intra– and subhepatic cysts is a single congenital disease with different types of dominance of one of the elements of the pathology of the ductal system.

99 years have passed since the first excision of the cyst of hepaticocholedochus McWhorter (1924). There are several approaches to surgical treatment: 1). Formation of biliodigestive anastomoses with the walls of a non-removed cyst, 2). Creation of hepaticoduodenoanastomosis [9]. Complete excision of cysts and imposition of hepaticoejunoanastomosis with the intestinal loop switched off according to Roux [1, 3, 5, 7, 13]. In publications, a significant percentage of stenoses of biliodigestive anastomoses and the development of malignancy are noted, complications are more common with non-removed fragments of cystic walls and cystodigestive anastomoses. S. C. Stain et al. (1995) revealed in 73% of patients stenosis after cystoenterostomy, which required surgery. The incidence of cancer was 26% [7].

2. Objective

To analyze the surgical treatment of patients with bile duct cysts, their complications and the choice of optimal methods of surgical correction depending on the localization of cysts.

3. Material and Methods

3.1. Material

Over the past 40 years, we have operated on 16 patients with cystic transformation of the bile ducts. There were 15 women and 1 man. The average age was 35 years. We use the classification of F. Alonso-Lej et al. (1959), dividing cysts by localization into 3 groups: hepatic, subhepatic and mixed [14]. In the subhepatic group (87%), we distinguish 3 subgroups: a) cysts of the lobar ducts and the place of their confluence, b) cysts of hepaticocholedochus, c) cysts of the retroduodenal department of choledochus. According to anatomical features, the localization of cysts was as follows: intrahepatic group – 1, lobular ducts and their fusion – 4, hepaticocholedoch – 9, distal choledochus – 1, mixed form – 1 (cyst of 2 and 3 segments of the liver (Caroli's disease) and hepaticocholedochus cyst).

3.2. Methods

The main diagnostic methods were ultrasound, endoscopic retrograde cholangiopancreatography, magnetic resonance cholangiopancreatography.

4. Results

The largest number of transformed cysts originated in the hepaticocholedochus (9), followed by – cysts of lobar ducts, and their fusion (4). Fusiform cysts were most common. Bile duct cysts were likely congenital in all 16 patients. It was found out from the anamnesis that in the first year of life (at 7 and 10 months) 2 girls were operated at a children's clinic for complicated cysts, one of which also had underdevelopment of the left hand. Subsequently, they underwent repeated interventions for cystic transformation of the bile ducts. The operation of choice in most cases was the excision of the walls of cysts and the imposition of a hepaticoejunoanastomosis with an isolated jejunum according to Roux (11). 2 patients with resection of both lobar ducts underwent hepaticoejunoanastomosis into the intestinal ring of an isolated jejunum (Figure 1) [15, 16]. Cystoenteroanastomosis was formed in 2 patients. Unresectable tumors of the subhepatic space were found in 2 patients 13 and 15 years after complex excision of cysts with hepaticoejunostomy.

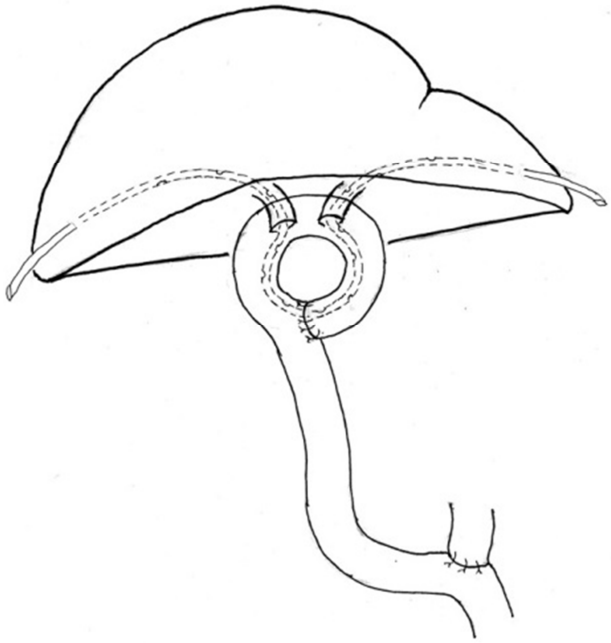

Figure 1. *Operation diagram. Bigepaticoejunoanastomosis into the intestinal ring of the isolated jejunum with bilateral replaceable transhepatic drainage.*

In 2 patients, invagination of fragments of the walls of cysts of the upper arch of the confluence of the lobar ducts into the common hepatic duct was revealed. In the available literature, we have not found publications about such a pathology. We give a detailed description of complex diagnostics, surgical findings and surgical techniques.

4.1. Clinical Observation 1

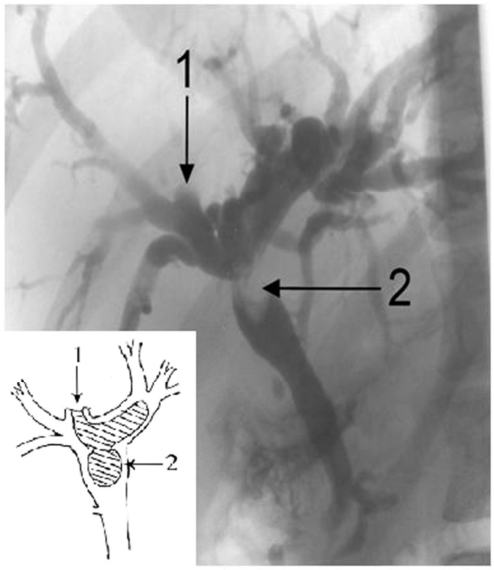

Figure 2. *Endoscopic retrograde cholangiopancreatography. Dilated hepatic ducts. The scheme of invagination of the cyst of the hepatic ducts.*

1. The contrasting area of the upper arch of confluence.
2. Irregularly shaped filling defects in the left lobar duct and terminal part of the common hepatic duct.

The 26–year-old patient had periodic pain in the liver, itching, periodic chills from the age of 9. Based on ultrasound data: chronic cholecystitis, a cyst of the liver gate measuring 50x40 mm of medium echo density associated with the portal vein; enlarged intrahepatic ducts (on the right – 14 mm, on the left – 12 mm). In July 1987, cholecystectomy and excision of parts of the cyst walls were performed. Thirty ml of bile was released through the safety drainage in 1 week. Histology: cyst of the liver ducts. 13 months after the operation, patient developed acute sharp pains in the liver and chills. After 5 days she was hospitalized in the clinic. On examination: temperature 37.6°; bilirubin – 92 mmol/L, increased alanine and aspartate transaminases. Ultrasound: Dilated intrahepatic ducts: right – 17 mm, left – 25 mm, choledoch – 8 mm. ERCP – intrahepatic ducts are dilated: right lobar – 17 mm, left – 25 mm with heterogeneous contrast. The common hepatic duct is up to 20 mm wide, in the proximal part contains a filling defect of 20x15 mm with uneven contours. On the upper surface of the confluence arch there is a rectangular 7x7 mm contrast area (Figure 2). Laparotomy was performed on 4.10.1988. The significant adhesive process of the liver gate area was divided. Enlarged hepatic ducts with thickened walls and features of inflammation were found. In the left lobe and common hepatic duct, tissue of medium density is palpated. Diagnosis: cancer of the hepatic ducts. We performed resection of lobular ducts and hepaticocholedoch with surrounding fiber. A bigepaticoejunoanastomosis was applied to the intestinal ring of an isolated jejunum with bilateral replaceable transhepatic drainage. The patient had an uneventful postoperative period and was subsequently observed for 4 years. Preparation: the lumen of the common hepatic duct and partially of the left lobar duct are filled with cystic tissue with bleeding, cherry-colored areas covered with fibrin. On the upper arch of the confluence, the mouth of a 7x7 mm cyst was found densely filled with an invaginated fragment of an extra-current cyst. Histology: cyst wall with fibrosis areas, mucosa contains cylindrical epithelium, macrophage infiltration. Conclusion – the wall of the cyst of the large bile duct.

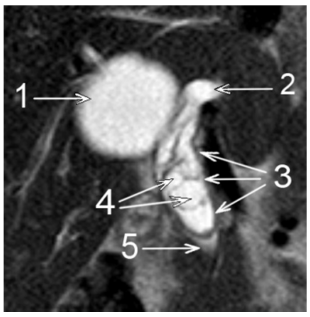

Figure 3. *Magnetic resonance cholangiopancreatography. Section 1. Extra ductal cyst and expanded common hepatic duct.*

1. Cyst of the area of the right lobar duct.
2. The distal part of the left lobar duct.
3. Expanded common hepatic duct.
4. Heterogeneous tissue in the lumen of the common hepatic duct.
5. The proximal part of the common bile duct.

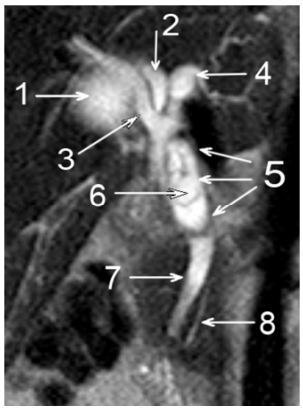

Figure 4. *Magnetic resonance cholangiopancreatography. Section 2. Ductal bile system.*

1. Extra ductal cyst.
2. The distal part of the cyst, emanating from the upper arch of the confluence.
3. Distal part of the right lobar duct.
4. Distal part of the left lobar duct.
5. Expanded common hepatic duct.
6. Heterogeneous density of tissue in the lumen of the common hepatic duct.
7. Common bile duct.
8. Distal part of the Virsung duct.

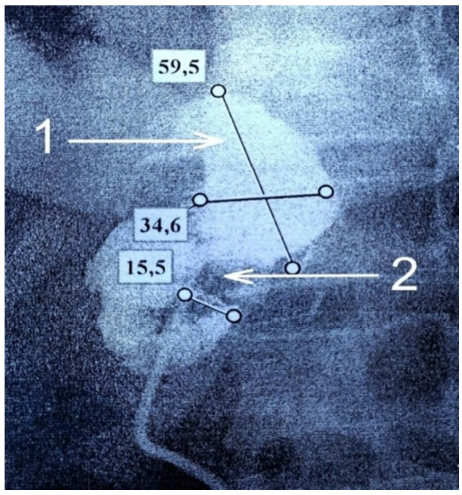

Figure 5. *Percutaneous cystocholangiography.*

1. Cyst.
2. Filling defects in the enlarged common hepatic duct.

4.2. Clinical Observation 2

The 51-year-old patient complained of intermittent pain in the liver, fever, icteric skin for 12 years. In 2011, laparotomy, cholecystectomy and partial excision of the cyst walls in the liver gate were performed. Histology: the walls of the cyst of the bile ducts. 100-200 ml of bile was released through the safety drainage for 2 months. After 2 years, patient experienced disease exacerbation. She was admitted to the clinic with the diagnosis of cyst of the liver gate. The MRCPG has been completed. In the area of the liver gate, a homogeneous structure is determined by a liquid formation with dimensions of 60x35x30 mm with uneven contours adjacent to the lobar ducts, expanded to 15 mm. The distal part of the cyst originates from the upper arch of the confluence. The common baked duct is expanded to 35 mm, 50 mm long, and contains a structure with areas of heterogeneous density in the lumen. The common bile and pancreatic ducts are not dilated (Figure 3, 4). Under ultrasound control, the cyst was drained with a bile flow rate of up to 300-400 ml. Percutaneous cystocholangiography was performed. The common hepatic duct is expanded to 30 mm, in the center there is an irregularly shaped filling defect of 15.5 x 20 mm (Figure 5). Clinical diagnosis: invagination of part of the cyst walls into the lumen of the common hepatic duct. Laparotomy was performed on 4.12.2013. Separation of a prominent cicatricial–adhesive process in the liver gate. A flabby cyst, intimately soldered to the dilated right hepatic duct, was revealed. In the general hepatic duct, an oblong, sedentary tissue of medium density was palpated. A further revision revealed an iatrogenic defect of 7x2 mm near the mouth of the wall of the left lobar duct, where part of the narrow wall of the extra-flow cyst is drawn in. Additional dissection of the duct wall in the area of the defect and hepaticotomy at the border with the stump of the cystic duct was performed. With difficulty, disinvagination of the cyst wall was carried out from the bottom up. All the walls of the extra-current cyst were excised. Transhepatic external drainage was carried out through the left lobar and cystic ducts. The bile flow was restored by suturing the walls of the dissected ducts. After 10 days, a transdrainage cholangiography was performed – the intrahepatic ducts decreased in diameter, the common hepatic duct had uneven walls, the contrast patency into the duodenum was timely. Patient had an uneventful postoperative period. She remained under observation for 5 years. Preparation: invaginate is covered with fibrin and indelible bile, walls with hyperemic areas. Histology: invaginate contains cyst walls with sclerosis phenomena, the mucosa is lined with prismatic epithelium with macrophage infiltration. Conclusion – the wall of the cyst of the large ducts of the liver.

5. Discussion

It is difficult to confidently explain the mechanism of invagination of fragments of the walls of cysts into the lumen of the ducts. As an invaginate, both transformed and biliary (true, simple) cyst walls with extrahepatic arrangement (on the leg) can act. It is impossible to exclude the participation of diverticula ducts. The process of invagination can be facilitated by a number of factors: 1. Complex topographic and anatomical relationships of lobar ducts, vessels and cysts. 2. Technical difficulties of excision of the walls of cysts, their partial removal. 3. Violation of fixation of fragments of the walls of cysts, and their mobility. 4. Iatrogenic defects of duct walls. 5. Defect of ligation of the walls of the mouth of the cyst. 6. The presence of a scar–adhesive process of the liver gate, isolating technical defects of the duct walls, due to the suction properties of the diaphragm, contributes to the retraction of a fragment of the cyst wall into the common hepatic duct. Given the high rates of malignancy of non-removed cysts, all their walls with surrounding fiber were excised in the presented clinical observations.

One of the causes of scarring of the biliodigestive anastomoses in cystic transformation of the bile ducts are congenital intra-current bridges of the distal lobe ducts. They create conditions for stagnation of bile, stone formation and infection in the ducts, structuring of anastomoses. Hepaticoscopy is recommended for diagnosis [5, 17]. H. Ando et al. (1995) reported that on the basis of surgical cholangiography, septal stenosis from the mucous and fibromuscular layers was detected in 80.6% of patients with cystic transformation of the bile ducts [17].

We found 2 cord-like cords in a 25-year-old patient in the distal part of the left lobe duct, which were excised before the imposition of the anastomosis. Another 23-year-old patient with a hepaticocholedoch cyst with dilated hepatic ducts underwent excision of the cyst walls, a bigepaticoejunoanastomosis was applied to the intestinal ring of an isolated jejunum without transhepatic drainage. After 4 years, the patient had a cholangitis clinic. MRCPG revealed stenosis of the left hepaticoejunoanastomosis with the expansion of the duct, in the distal part of which cord-shaped bridges and a large oblong concretion were determined (Figure 6). Relaparotomy operation (September 2022).

Resection of the anastomosis, 20 mm duct with lintels and concretion, repeated reconstruction of the anastomosis with transhepatic drainage was performed. Smooth postoperative period. The patient is under observation.

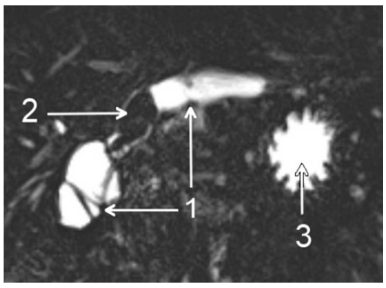

Figure 6. *Magnetic resonance cholangiography.*

Stenosed, dilated left lobar duct.
1. Intra-current jumpers of the left lobe duct.
2. A large oblong stone in the duct.
3. Stomach.

6. Conclusion

Cystic transformation of the bile ducts is rare and typically affects large bile ducts. It lacks specific and pathognomonic symptoms, but often leads to malignancies of the walls of cysts and stenosis of cystodigestive anastomoses. The optimal diagnostic method is MRCPG. All walls of cysts should be completely removed with the imposition of hepaticoejunoanastomosis according to Roux. The mouths of the lobe ducts must be examined with the help of MRCP for the presence of intra-flow bridges that are subject to excision. In subhepatic cysts, the distal sections of the dilated lobar ducts are recommended to be resected with the formation of a bigepaticoejunoanastomosis into the intestinal ring of the isolated jejunum using a seromuscular single–row continuous suture with absorbable threads 4/0-5/0. An individual approach is needed in the treatment of invaginated fragments of the walls of cysts in the common hepatic duct: disinvagination, resection of the hepatic ducts. In case of single lobar cystic transformations of cysts, a resection technique is shown.

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Participation of Authors

Concept and design of the study – G.D., I.D.
Collection and processing of material – G.D., E.K., S.B., I.D., S.S.
Statistical processing – I.D., S.B., E.K., S.S.
Text writing – G.D., I.D.
Editing – G.D., I.D.

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