ISSN 2181-3187

UDC- 616.36 – 981.42 – 089 - 008.5 – 002.64

ECHINOCOCCOSIS OF THE LIVER COMPLICATED BY A BREAKTHROUGH INTO THE BILIARY TRACT AND THE DEVELOPMENT OF MECHANICAL JAUNDICE: DIAGNOSIS AND TREATMENT (LITERATURE REVIEW)

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Resume.

Hepatic echinococcosis is a serious medical issue, especially in endemic regions. Complications of the disease, such as cyst rupture into the bile ducts leading to the development of mechanical jaundice, occur in 10-30% of patients and significantly worsen the clinical course, resulting in cholangitis, liver abscesses, and liver failure. Diagnosing these conditions requires the use of high-precision imaging techniques such as ultrasound, MRCP, and ERCP. The rupture of an echinococcal cyst into the bile ducts necessitates a multidisciplinary approach, including endoscopic removal of parasitic elements and surgical intervention, followed by bile duct drainage. Modern techniques, such as minimally invasive endoscopic procedures, reduce the risk of postoperative complications and improve treatment outcomes.

ISSN 2181-3187

The relevance of the issue lies in the high frequency of complications and diagnostic challenges, which necessitates further improvement of treatment methods and the implementation of minimally invasive technologies.

Key words: Echinococcosis of the liver, mechanical jaundice, cyst breakthrough into the bile ducts, cystobiliary fistulas, endoscopy, complications.

Relevance

Hepatic echinococcosis represents a serious medical challenge, particularly in regions with a high endemicity of the disease. In recent years, increased attention has been paid to complicated forms of echinococcosis, such as cyst rupture into the bile ducts, which can lead to the development of mechanical jaundice and, in prolonged cases, hepatic failure. These complications significantly worsen the clinical course of the disease and complicate both diagnosis and treatment [4, 5, 7, 11, 14, 20, 25, 35].

The relevance of the problem lies in the limited effectiveness of conservative treatment at advanced stages of the disease. According to the World Health Organization (WHO), the prevalence of echinococcosis in endemic areas remains high, necessitating the development of new diagnostic and therapeutic approaches aimed at reducing the incidence of complications and improving patient outcomes [8, 14, 21, 25, 29, 30, 32, 37, 55].

In endemic regions such as Central Asia, South America, and parts of Europe, echinococcosis remains a significant medical issue. Complications such as cyst rupture into the bile ducts and the development of mechanical jaundice are observed in 10–30% of patients and present complex clinical challenges. These complications require timely diagnosis and a multifactorial therapeutic approach [29, 30, 37, 38, 41, 44, 45, 55, 58].

The rupture of an echinococcal cyst into the bile ducts leads to severe complications such as mechanical jaundice, cholangitis, and liver abscesses. Approximately 20–30% of patients with hepatic echinococcosis develop mechanical jaundice due to a high level of parasitic obstruction of the biliary tract, which often

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requires surgical intervention. Lack of early diagnosis and treatment increases mortality and the risk of severe complications [1, 7, 9, 10, 11, 12, 16, 23, 34].

According to various authors, there are often fistulous connections of various sizes between the cyst and adjacent bile ducts, referred to as cystobiliary fistulas. These fistulas, which result from a silent rupture of the cyst into the bile ducts, have been identified in up to 90% of patients with hepatic echinococcosis according to some studies [15, 25, 41].

Pathogenesis. The rupture of a hepatic echinococcal cyst into the bile ducts can, in rare cases, lead to the development of anaphylactic shock, although the more common complication is the onset of mechanical jaundice.

Hepatic echinococcosis complicated by cyst rupture into the bile ducts poses a serious threat to the patient's life and represents a significant clinical challenge due to difficulties in timely diagnosis and management. According to Mohamed et al. (2022), the incidence of cyst rupture into the bile ducts ranges from 10% to 37%, leading to jaundice in the majority of patients. Diagnostic delays may result in complications such as cholangitis, liver abscesses, peritonitis, chronic fistulous processes, and hepatic failure, all of which negatively affect treatment outcomes [38].

The critical nature of this problem is also related to insufficient awareness among medical professionals. Hepatic echinococcosis complicated by mechanical jaundice often presents with symptoms that mimic other conditions, such as cholelithiasis or pancreatic tumors, making differential diagnosis essential.

The rupture of daughter cysts or parasitic elements into the hepatic duct (hepaticholedochus) can lead to biliary obstruction, which in turn causes mechanical jaundice and cholangitis [50].

According to Sh. Sh. Amonov et al., the development and frequency of complications during surgical treatment of hepatic echinococcosis involving the bile ducts depend on multiple factors: the duration of the disease, size and location of the cysts, presence of cyst complications (e.g., suppuration or calcification), the nature of

ISSN 2181-3187

biliary involvement, and the chosen surgical approach. The same authors report that biliary fistulas persist postoperatively in 12% of cases [5].

The rupture of the contents of an echinococcal liver cyst into the bile ducts is considered the second most common complication after suppuration and is among the most severe in the course of this disease [16, 25, 40, 41].

The size and location of the parasitic cyst are key risk factors for the development of communication between the cyst cavity and the biliary ducts. When the cyst diameter exceeds 7.5 cm, the likelihood of forming a cystobiliary fistula reaches up to 79% [11, 19, 21].

Clinical Manifestations

The clinical presentation of hepatic echinococcosis, including involvement of the biliary tract, can range from mild or subclinical symptoms—as seen with cystobiliary fistulas—to severe manifestations such as obstructive jaundice and cholangitis. Rupture of an echinococcal cyst into the bile ducts is usually accompanied by the sudden onset of mechanical jaundice, fever, and pain in the right upper quadrant. In such cases, itching, dark-colored urine, and pale stools, which are typical of biliary obstruction, become diagnostically significant.

The symptoms vary depending on the nature and extent of biliary tract involvement. The most serious complication of cyst rupture into the bile ducts is cholangitis. Surgical treatment of echinococcosis complicated by biliary fistulas is technically demanding, as surgeries are often performed under critical conditions, including septic cholangitis, obstructive jaundice, severe intoxication, and hepatic failure. This results in high rates of postoperative complications and a mortality rate reported between 25% and 47% in different studies. The variability is due to some authors including only large fistulas in their statistics, while others also count smaller ones [4].

As the parasite grows, all components of the cyst enlarge, leading to increased intracystic pressure and stretching of the capsule. Small biliary fistulas in the fibrous capsule expand, and new fistulas may form as cracks develop in the bile duct wall. The

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size of the perforation opening can vary from a few millimeters to several centimeters. Typically, one opening forms, though multiple perforations are less common.

A large or suppurated echinococcal cyst located near the main bile ducts can compress them, resulting in obstructive jaundice, or it may rupture into the ducts. In the latter case, dead or viable daughter cysts, along with fragments of the chitinous membrane, can obstruct the ducts, leading to severe infectious-allergic cholangitis, hepatic failure, or, in some cases, acute cholangiogenic pancreatitis.

Rupture of a hepatic echinococcal cyst into the bile ducts is typically diagnosed intraoperatively when, after evacuating the cyst contents, a bile-leaking orifice is found on the inner surface of the fibrous capsule. The margins of the opening are dark green, and bile continues to ooze even after thorough blotting with gauze. Persistent biliary fistulas in the postoperative period are observed in approximately 12% of cases [4].

Based on their experience with 2,785 patients with hepatic echinococcosis, X. Wu et al. reported that 37 patients (1.3%) had confirmed cyst rupture into the bile ducts. The authors suggest that in endemic areas, complaints of right upper quadrant pain or heaviness, together with signs of jaundice, should raise suspicion of cyst rupture into the biliary tract. This diagnosis should be confirmed using ultrasound imaging and computed tomography [33, 56].

Diagnosis

Laboratory diagnostic methods.

In patients with hepatic echinococcosis complicated by jaundice, laboratory findings typically include elevated serum bilirubin levels, as well as increased activity of liver enzymes, particularly alkaline phosphatase (ALP) and gamma-glutamyl transferase (GGT).

Imaging and instrumental diagnostics.

The primary diagnostic tools remain ultrasound (US) and magnetic resonance cholangiopancreatography (MRCP). These methods not only help identify cysts, but also assess the degree of bile duct involvement and detect fistulous tracts. In addition, endoscopic retrograde cholangiopancreatography (ERCP) is used to detect fistulas and

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evaluate the feasibility of drainage.

ERCP is considered the most effective method for preoperative diagnosis of cystobiliary fistulas. It also allows for sphincterotomy and decompression of the common bile duct (CBD) if needed. ERCP plays a key role in diagnosing hepatic echinococcosis with biliary involvement. When combined with nasobiliary drainage, ERCP helps stabilize the patient's condition prior to surgery and facilitates operative planning at the optimal time [13, 23, 28].

MRI with cholangiopancreatographic sequencing enables highly accurate detection of biliary fistulas, which is especially important in planning minimally invasive interventions. MRCP provides clear visualization of biliary anatomy and allows for precise localization of obstructions [8, 24, 29, 51].

Ultrasound (US) remains the main diagnostic method for hepatic echinococcosis due to its availability, noninvasiveness, and high informativeness. On ultrasound, signs include dilation of bile ducts with hyperechoic inclusions inside, representing cyst contents such as chitin fragments and daughter cysts. In cases of significant rupture, cyst elements may enter the gallbladder, appearing as echogenic material within the gallbladder lumen. In some cases, gas in the cyst cavity, detached chitin membrane, cystobiliary fistulas, and round fluid collections in the bile ducts may also be detected [39].

Obstruction of the bile ducts by dense cyst fragments occurs in 5–10% of cases and leads to mechanical jaundice. This condition is commonly seen when the cystobiliary fistula has a diameter of at least 5 mm. To resolve biliary leakage from residual cavities, endoscopic techniques are employed, such as endoscopic sphincterotomy with duodenobiliary stenting or nasobiliary drainage [27, 42, 52].

Biliary tract involvement in hepatic echinococcosis is associated with a severe clinical course, frequent diagnostic errors, and a persistently high mortality rate. Under these circumstances, it is particularly important to continue studying the clinical characteristics and patterns of biliary complications associated with this disease.

Treatment

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There is still no unified consensus among surgeons regarding the extent of surgical intervention required for hepatic echinococcosis with biliary tract involvement. Some specialists advocate for an individualized and case-specific surgical approach, depending on the anatomical and pathological characteristics of each case [4, 20, 43, 58].

A widely accepted approach for internal cystobiliary fistulas is their closure from within the fibrous capsule using atraumatic sutures. In cases of cyst rupture into the bile ducts accompanied by mechanical jaundice and cholangitis, many experts recommend choledochotomy to remove parasitic elements from the ducts, followed by external drainage using a T-tube [27, 35, 52].

The implementation of minimally invasive techniques in the surgical treatment of biliary fistulas is a promising direction, as it may improve liver function recovery, disease prognosis, and quality of life for patients [4].

In patients with hepatic echinococcosis complicated by rupture into the bile ducts and mechanical jaundice with cholangitis, choledochotomy with T-tube drainage remains the preferred method to remove parasitic elements and restore biliary outflow.

Surgical management is often complicated by undiagnosed intraoperative cystobiliary fistulas, which can result in persistent postoperative bile leakage and the development of external biliary fistulas. External drainage of the residual cavity in cases of cystobiliary fistula carries a high risk of complications, such as bile fistula formation and cavity suppuration, significantly prolonging postoperative recovery [35, 52].

Aliyev M.A. et al. reported successful use of laparoscopic techniques to manage a case where a cystobiliary fistula was detected intraoperatively. Following coagulation of the fistula, drainage of the residual cyst cavity, cholecystectomy, and Pikovsky biliary drainage were performed. No postoperative complications occurred [2, 3].

According to Seisembayev M.A. et al., when closure of the fistula from within the fibrous capsule is technically impossible or inappropriate (e.g., with large fistulas involving lobar ducts), cholecystectomy with T-tube choledochal drainage is indicated.

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One limb of the drain is inserted into the fistulous right hepatic duct, and omentoplasty is used to obliterate the residual cavity and prevent the development of an “undrainable lobe” syndrome [10].

Ilkhamov F.A. et al. (1998) propose a three-stage treatment strategy for severe jaundice-associated forms:

1. Preoperative biliary decompression using EPST and nasobiliary drainage,
2. Echinococcectomy with closure of the biliary fistula and management of the residual cavity,
3. Postoperative therapy to control cholangitis, hepatic insufficiency, and promote liver regeneration [1, 17, 27].

External Biliary Fistulas

One of the main challenges in treating hepatic echinococcosis complicated by rupture into the biliary tree is that patients often present at the hospital with varying stages of hepatic failure due to mechanical obstruction and biliary hypertension. This significantly worsens their condition, increases the risk of postoperative complications and recurrence, and prolongs the period of postoperative rehabilitation. Preoperative biliary decompression techniques for ruptured parasitic cysts and endoscopic interventions for managing external biliary fistulas—which commonly arise after echinococcectomy—have received insufficient attention [14, 26, 30, 44].

Postoperative complications, including external biliary fistulas, residual cavity infections in the liver, and mechanical jaundice (in 10–24% of cases), are often due to unidentified or untreated cystobiliary fistulas during surgery [12].

Postoperative bile leakage and fistula formation are most commonly caused by cystobiliary communication (CBC) due to intrabiliary rupture (IBR) of the cyst—this being the most frequent complication after hepatic echinococcal surgery. The incidence of postoperative bile leakage varies between 2.5% and 28.6%. If the leak persists for more than 10 days and drains externally, it is classified as a biliary fistula, which is reported in 1–25% of cases and is a major source of postoperative morbidity [22, 46].

In the absence of proper internal or external drainage, complications such as

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biliomas, biliary abscesses, or biliary peritonitis may develop, potentially progressing to sepsis and resulting in severe morbidity or mortality. Postoperative biliary fistulas are a major cause of prolonged hospitalization and interventions following echinococcectomy. However, some fistulas may close spontaneously within the first postoperative week. They are categorized as low-output fistulas if the drainage volume is <300 mL/day or high-output if >300 mL/day [22].

If a fistula persists for more than 3 weeks or is high-output, endoscopic or surgical intervention should be considered instead of conservative treatment [25, 41].

The most frequent complication of cystic echinococcosis (CE) is cystobiliary communication, found in approximately 60% of complicated cases. Two main pathogenetic theories have been proposed:

Progressive necrosis of the bile duct wall due to compression by the echinococcal cyst, leading to the formation of a cystobiliary communication.

Atrophy and rupture of small biliary radicals that penetrate the pericystic membrane and undergo pressure-induced damage [41].

In cases of postoperative external fistulas, Vagianos C. et al. described a patient who underwent echinococcectomy for a giant hepatic cyst complicated by an external biliary fistula. Endoscopic papillotomy (EPST) combined with nasobiliary drainage and continuous bile aspiration led to successful fistula closure in a short time [53, 54].

Most authors agree that eliminating the root cause—biliary hypertension—is key in the treatment of external biliary fistulas. Tekant Y. et al. reported successful closure of the fistula in 9 out of 10 patients after EPST within 2 to 15 days post-procedure [49].

Various approaches have been suggested for managing post-echinococcectomy biliary fistulas, including:

Embolization of the distal fistula tract using 1.5 mL of histoacryl, biliary stenting, and Percutaneous embolization of biliary fistulas [18, 31, 34, 57].

A review of the literature highlights that surgical treatment of hepatic echinococcosis complicated by biliary tract involvement remains a significant challenge, primarily due to the difficulty of diagnosing biliary injury and the lack of a

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standardized surgical approach. These issues contribute to a high rate of postoperative complications and elevated mortality.

In cases of echinococcal cyst rupture into the bile ducts, the primary strategy involves endoscopic removal of chitin membranes from the ducts, followed—if possible—by echinococcectomy.

However, it remains unclear whether cystobiliary fistulas can be completely closed in the postoperative period, and which methods are most effective. Furthermore, the effects of germicidal agents used to treat residual cavities on the biliary tract in the presence of fistulas have not been sufficiently studied. The impact of various agents on fibrous tissue, and their comparative antiparasitic and toxic properties, have not been adequately characterized.

The surgical treatment of hepatic echinococcosis complicated by biliary tract injury remains a major unresolved issue due to diagnostic difficulties and the absence of unified surgical protocols. This results in a high rate of postoperative complications and significant mortality.

Pharmacological Therapy

Pharmacological treatment includes the use of antiparasitic agents such as albendazole and mebendazole to reduce the risk of recurrence and prevent further growth of hydatid cysts. Antiparasitic therapy is typically prescribed for a prolonged period after surgery to eliminate residual parasitic elements and minimize the chance of recurrence.

Among antiparasitic medications, albendazole remains a cornerstone of post-surgical treatment, aimed at preventing disease recurrence and destroying any remaining parasitic components. It is usually prescribed at a dose of 10–15 mg/kg/day, continued for 3 to 6 months after the surgical procedure.

This regimen plays a critical role in long-term disease control and is particularly important in patients with incomplete cyst removal, intraoperative cyst spillage, or multiple organ involvement.

Unresolved Issues in Treatment

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1. Endoscopic Techniques and Incomplete Parasite Removal

Endoscopic methods such as ERCP and EPST with removal of parasitic material are commonly employed to restore bile flow and reduce the risk of cholangitis. However, these procedures may be insufficient for complete removal of all cyst elements, which increases the risk of recurrence. Therefore, there remains an unresolved issue regarding the endoscopic delivery of antiparasitic agents directly into the biliary tract or residual cavity to reduce recurrence rates.

2. Management of High-Risk Patients (e.g., Liver Cirrhosis).In high-risk patients, particularly those with liver cirrhosis, the choice of treatment becomes more complex. The decision between surgical intervention and minimally invasive procedures remains unresolved, especially in complicated or borderline clinical scenarios. There is a pressing need to develop and validate preferred treatment algorithms for such cases.

3. Post-Intervention Pharmacotherapy. A key unresolved question pertains to the optimal duration and regimen of antiparasitic therapy (e.g., albendazole or mebendazole) following surgical or endoscopic interventions. The long-term safety, efficacy, and toxicity of these drugs, especially in patients with impaired liver function, need further clarification. Careful consideration of dose adjustments, monitoring of liver enzymes, and management of side effects is essential.

Conclusion

Hepatic echinococcosis complicated by rupture into the bile ducts and parasitic mechanical jaundice represents a complex clinical challenge that requires a multidisciplinary approach to diagnosis and treatment. Rupture of an echinococcal cyst into the bile ducts is a serious complication of hepatic echinococcosis, frequently associated with mechanical jaundice, cholangitis, and hepatic insufficiency. The diagnosis of this condition is often difficult due to nonspecific clinical manifestations and the need for advanced imaging techniques such as MRI and ERCP, which may not always be available in endemic regions.

Effective treatment requires a combination of surgical and endoscopic

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interventions aimed at biliary decompression and prevention of recurrence. Endoscopic methods, such as ERCP with extraction of parasitic material and bile duct decompression, are widely used; however, their efficacy is limited in cases involving large fistulas or complex cysts.

The optimal approach to managing cystobiliary fistulas and preventing recurrence remains unresolved. Long-term antiparasitic therapy (e.g., with albendazole) following surgery is recommended to prevent recurrence, but its efficacy and safety require further study, especially in patients with comorbidities such as liver cirrhosis.

Further research is needed to optimize diagnostic and therapeutic strategies, develop more effective minimally invasive techniques, and establish evidence-based antiparasitic treatment protocols to improve outcomes in patients with complicated hepatic echinococcosis.

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