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Case Report

Total vascular isolation in segmental liver resection for echinococcal cyst: case report

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ABSTRACT

Liver echinococcosis is a widespread parasitic disease that does not tend to decrease in endemic regions. High frequency of complications, reaching up to 57%, relapses of the disease, accompanied by repeated surgeries, leave the problem relevant at the present time. The main purpose of the work is to improve the results of treatment of patients with echinococcal liver cysts. This article presents, the outcomes of surgical interventions in two patients, with echinococcal liver cysts who underwent segmental liver resections of the liver, employing total vascular isolation (TVI) technique, which is carried out at the department of pancreatology hepatology and transplantation of organs and tissues, Grodno regional clinical hospital in Grodno, Belarus. Our entire team performed segmental liver resection of echinococcal cysts using the technique of TVI for the both patients who presented with symptoms of abdominal discomfort in their right upper quadrant and further imaging studies confirmed the presence of cystic lesions within the 6 and 7 liver segments, intricately intertwined with major hepatic vessels. Both the patients had optimal surgical outcomes with minimal intraoperative blood loss while preventing major complications including air embolism, with the employment of the method of TVI. The use of the method of TVI allows to significantly reduce intraoperative blood loss, and also makes it possible to avoid extensive resections in echinococcal liver cysts.

Keywords: Liver echinococcosis, Surgical treatment, Liver resection, Vascular isolation

INTRODUCTION

Echinococcosis is a widespread parasitic disease across the globe. According to some estimates, more than 1 million people in the world are currently affected by echinococcosis, and the incidence in some endemic and non-endemic regions varies more than 200 times. In the last decade, there has been an increase in the incidence of echinococcosis and the expansion of the geographical boundaries of the disease.[1]

Echinococcosis is caused by the parasitic tapeworm of genus *Echinococcus*. The two most important forms which infect the human are; *Echinococcus granulosus* which produces unilocular cystic lesions of the liver and *Echinococcus multilocularis* which produces multilocular alveolar cysts in the lungs.[2]

Echinococcal species have both definitive (dogs) and intermediate (sheep, human) hosts. Eggs are passed in the feces of definitive hosts and they are ingested by the intermediate hosts. Likewise, humans get infected as an intermediate host by ingestion of water, wild berries or plants which are contaminated with parasitic eggs or by having direct contact with the definitive hosts. After ingestion of eggs, embryos are escaped from the eggs, penetrate the intestinal mucosa, gain access to the mesenteric vessels, thereby enter the portal circulation and get carried to various organs.[2] Liver is the most commonly affected organ while lungs are the second most common location and rarely can present in virtually

any other organ.[3] The risk factors associated with human infection of echinococcosis include uncontrolled dewormed dogs raising at home, uncontrolled livestock slaughter and unhygienic living conditions.

This article solely emphasizes on cystic echinoccosis of liver which is caused by *Echinococcus granulosus* and is characterized by the development of parasitic cysts.[4] The cysts usually range from 1-15 cm in diameter and its growth ranges from 1-2 mm to 10 mm per year. The cysts prone to affect the right lobe of the liver more frequently than the left lobe due to the diversity of portal blood flow.[5] Echinococcal cysts consist of a peri parasitic host tissue and it surround the larval endocyst and an endocyst itself. The endocyst consist of an outer laminated layer and an inner germinal layer. Germinal layer gives rise to brood capsules (germinating cysts) and daughter cysts. Brood capsules rupture and release viable protoscoleces (future heads of the adult worms) which look like white sediments. They float inside the cyst, so they are called as “hydatid” sand. The cyst is filled with clear fluid in early stages but can be turbid and yellowish, with fragments of endocysts in advanced stages.[3]

Echinococcosis of the liver usually remain asymptomatic during the early stages until the cysts expand gradually and affect the liver tissue and elicit pressure symptoms. As the cysts expand, depending on the location and size of the tumor, they exert pressure on nearby structures letting the patient feel abdominal pain, discomfort in the right upper quadrant along with nausea, vomiting and poor appetite. And physical findings include hepatomegaly, abnormal palpable mass in the right upper quadrant and abdominal distention. These growing cysts also may compress the surrounding structures. For instance; cysts can compress the bile duct leading to its obstruction, which can finally manifest as obstructive jaundice. Furthermore, rupture of the cysts may produce acute hypersensitivity reactions elevating the levels of IgE, IgG manifesting fever, pruritus, eosinophilia or even fatal anaphylaxis.[2,3,5]

As early-stage liver cystic echinococcosis (CE) is often asymptomatic, diagnosis is frequently incidental, and it's often discovered during abdominal ultrasound performed for unrelated reasons. In endemic regions, symptoms coupled with exposure to sheepdogs raise suspicion of CE. Diagnosis typically combines patient's clinical history, imaging, serological tests and ultrasound being the gold standard for assessing cyst number, location, size, and viability though MRI offers superior visualization of internal cyst structures and is useful pre-surgically. While ultrasound is excellent for characterizing cysts, additional imaging (MRI or CT) may be needed to differentiate CE from other lesions. Immunodiagnostic tests confirm imaging findings and monitor treatment response, although sensitivity varies depending on cyst integrity with ruptured or leaking cysts generating stronger immune responses.[5,6]

The first discoverers of echinococcus are the doctors of ancient Greece. Hippocrates described the liver damage as a "liver filled with water".[4] Scientists of that time misunderstood echinococcal cysts as expansion of lymphatic vessels and called them "hydatids".[4] Rudi (1681) first spoke about the parasitic nature of echinococcosis. Thornton successfully removed an echinococcal liver cyst for the first time in 1883. In recent years, despite the widespread introduction of highly informative examination methods and high-tech methods of surgical treatment of patients with liver echinococcosis, their number continues to increase, moreover, there is a noticeable increase in the frequency of its complicated forms and recurrence of the disease. The high frequency of postoperative complications, reaching up to 57%, relapses of the disease, occurring in the range from 3 to 54%, are accompanied by repeated surgeries, leave the problem of treatment of this pathology relevant.[7] According to many researchers, one of the factors significantly affecting the effectiveness of treatment is the choice of surgical intervention method.[1] Radical operations such as peri-cystectomy, liver resection, right-sided and left-sided hemihepatectomy remain the main methods of surgical treatment, despite their traumatic nature and high risk of intraoperative bleeding. According to Reifferscheid the liver is the topographical center of the most complex vascular systems.[8]

Therefore, the most dangerous intraoperative complication is bleeding, which in some cases can be fatal. Due to the above reason, reducing the degree of blood loss is one of the priorities in liver surgery, especially with the intimate location of focal formations with liver vessels.

The study of the segmental structure of the liver and the concept of controlled hemi-hepatectomy, along with the method of Pringle, made it possible to significantly reduce the risk of hepatectomy, but the achievements did not effect the resections of performed formations in the field of confluence of hepatic vein and inferior vena cava.[9] Even under the conditions of using Pringle, damage to large hepatic veins leads to massive blood loss as a result of retrograde blood flow from the inferior vena cava and is fraught with air embolism.[9] In 1974, J. Fortner et al, for the prevention of bleeding in extensive resections, a method of complete vascular isolation and hypothermic perfusion of the liver was proposed.[8] A simpler and more commonly used technique is the modified technique of general hepatic occlusion without perfusion of hypothermic solutions. The method of TVI consists of compression of the hepatoduodenal ligament and inferior vena cava in its supra- and sub hepatic parts. According to H. Bismuthetal which describe the experience of 51 extensive liver resections using the method of complete vascular isolation, its duration was from 20 to 90 minutes, while the volume of intraoperative blood loss decreased by 2-3 times.[8,10]

Purpose of the study was to improve the results of surgical treatment of patients with echinococcal liver cysts. In this article, we present the results of surgical treatment of two patients who underwent segmental liver resection using the technique of TVI for echinococcal liver cyst.

CASE REPORT

A 56-year-old female patient and a 45-year-old male patient were admitted to the department of surgical pancreatology, hepatology and transplantation of Grodno Regional clinical hospital, Belarus with complaints of heaviness and aching pain in the right upper quadrant of the abdomen with objective examination. The general condition of patients is satisfactory; the skin is pale pink. The abdomen is soft, slightly painful in the right upper quadrant of the abdomen. Both patients underwent a complex of laboratory and instrumental research methods, including MRI, CT and ultrasound of the abdominal and retroperitoneal organs. According to liver ultrasound, both patients in the 7th segment of liver visualized liquid formations, most likely of parasitic origin. In the first case, 48×30 mm with a parietal tissue component of 13×13 mm and the presence of an inflammation zone. In the second case, 40×31 mm cm also with the presence of a parietal component. According to MRI data (Figure 1), both patients in the S7 segment of the liver subcapsularly revealed rounded focal formations with clear contours of 50×33 mm and respectively 45×30 mm, with the presence along the posterior wall of a hyperdensive component with clear contours of 5×6 mm. The fact of the intimate location of liquid formations to the right hepatic vein in both cases has been established. No pathologies on the part of other organs were detected.

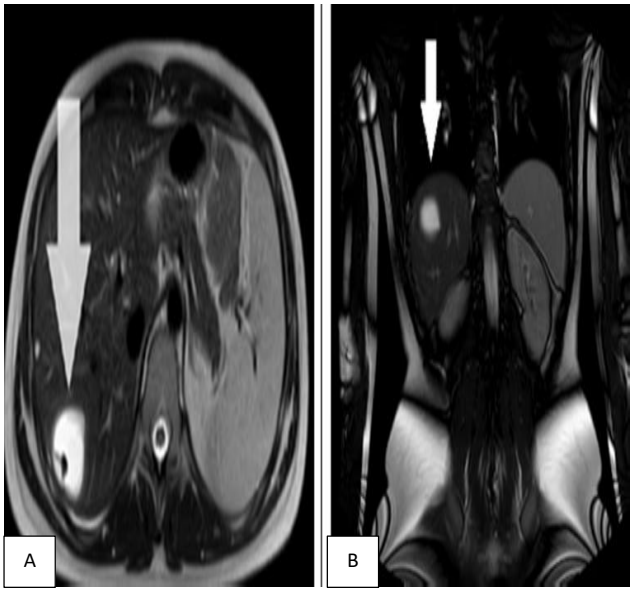

Figure 1 (A and B): MRI-scan of the abdominal cavity.
Arrows indicate focal formations in the right lobe of the liver.

The results of immune-enzyme analysis for echinococcal antigens were positive in both cases.

Based on the history, complaints, objective examination, laboratory and instrumental studies, the following clinical diagnosis was made.

Echinococcal cyst in the 7th segment (S7) of the right lobe of the liver.

Patients underwent segmental resection of the liver for an echinococcal cyst using the method of TVI. In both cases, a laparotomy was performed with a J-shaped approach in the right upper quadrant of the abdomen. During the revision of the abdominal cavity, no effusion was found. During palpation in the seventh segment of the liver, the volumetric formation of 5×3 cm and, accordingly, 4×3 cm, was palpated by fluctuation and uneven contours, located in a capsule of whitish color - most likely an echinococcal cyst (Figure 2).

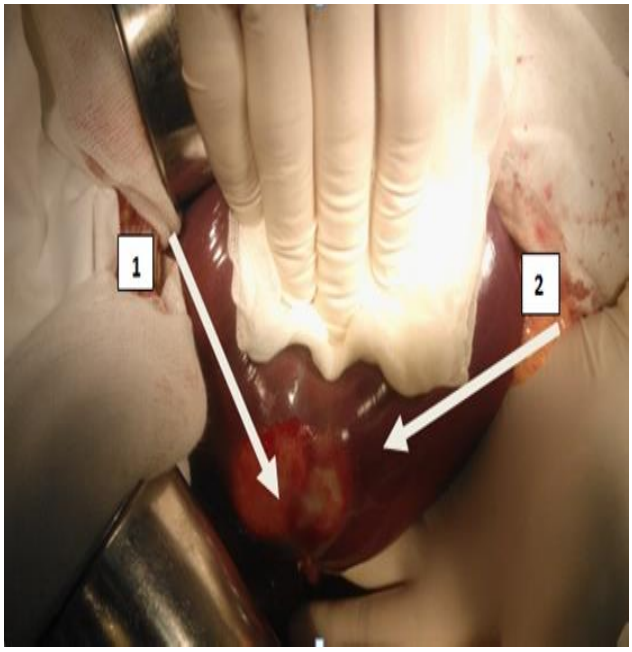

Figure 2: Echinococcal cyst in S7 of the right lobe of the liver.
1-Echinococcal cyst and 2-the right lobe of the liver.

Despite the presence of MRI of signs of echinococcal cyst invasion into the right hepatic vein, it was decided to attempt to perform resection of the seventh segment of the liver using TVI to minimize intraoperative blood loss in case of damage to the right hepatic vein during the mobilization of echinococcal cyst.

The right triangular and coronary ligaments of the liver were clamped. To carry out TVI of the liver, the inferior vena cava in the supra- and subhepatic sections, as well as the hepatoduodenal ligament were mobilized using a thread-Pringle maneuver (Figure 3 and 4) The thread was held upwards in case of bleeding develops.

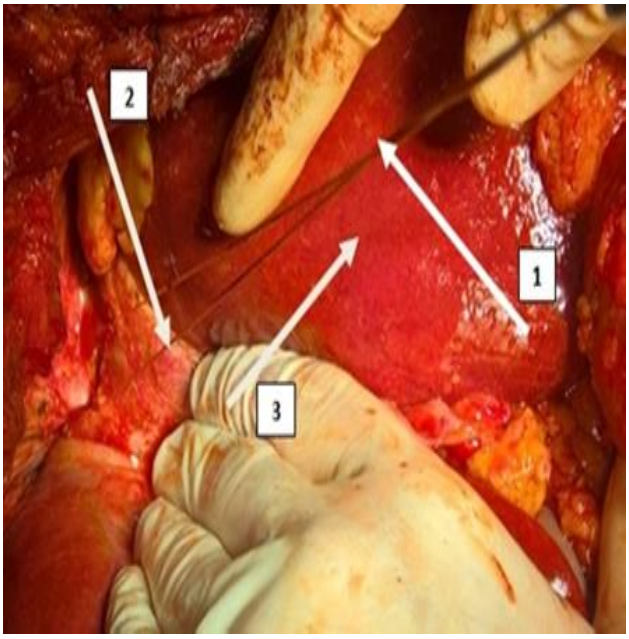

Figure 3: Control (safety) ligature on the supra-hepatic part of the inferior vena cava.
1-Ligature, 2-the suprahepatic department of the inferior vena cava and 3-the left lobe of the liver.

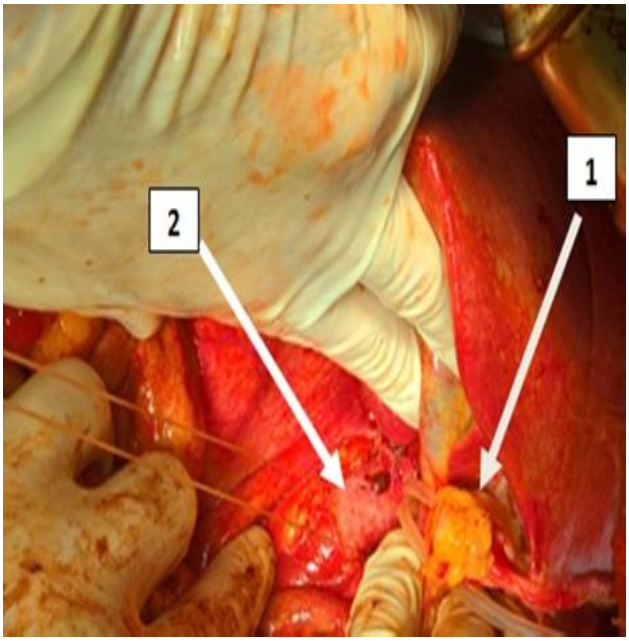

Figure 4: Control (safety) ligature on the subhepatic part of the inferior vena cava. Pringle maneuver.
1-Hepato-duodenal ligament and 2-the subhepatic part of the inferior vena cava.

A trial compression of the inferior vena cava was performed-the hemodynamics was stable, the systolic blood pressure decreased to 105 mmHg.

Along the perimeter of the echinococcal cysts of the 7th segment on the liver capsule, the resection boundary was outlined by 1cm from the cyst boundaries (Figure 5).

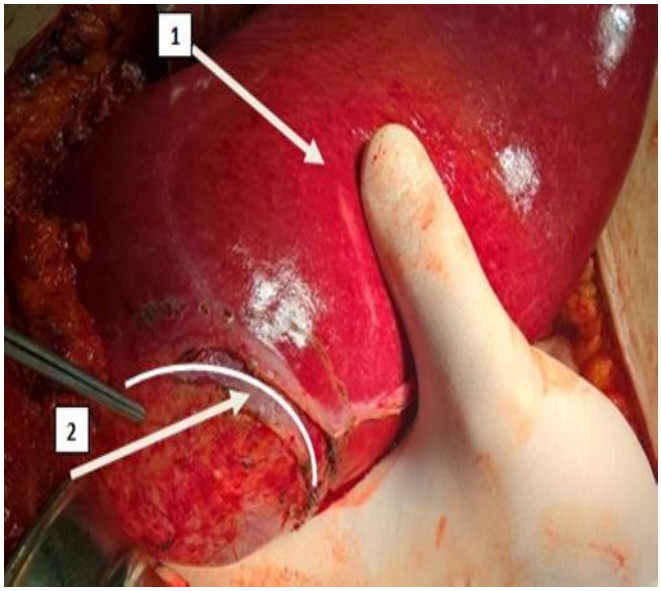

Figure 5: The boundaries of resection of the seventh segment of the liver with a cyst.
1-The right lobe of the liver and 2-the boundaries of resection.

Next, 6 and 7 segments of the liver were mobilized from the inferior vena cava with the lower hepatic veins. During the mobilization of one of the veins, one patient was found to have a linear traction defect in the wall of the inferior vena cava. Thanks to the use of TVI, intraoperative blood loss was avoided at this stage. The place of "linear traction " was stitched with a vascular suture over the vascular clamp of Satinsky. Liver parenchyma transsection was performed using ligasure triad apparatus, bipolar and monopolar coagulation, as well as using precision stitching of visualized vascular and biliary structures. Resections of the 7th segment of the liver with the above-described formations were performed step by step. At the same time, with great technical difficulties, it was possible to separate the cyst from the intimately soldered right hepatic vein and its branches to 6 and segments (Figure 6).

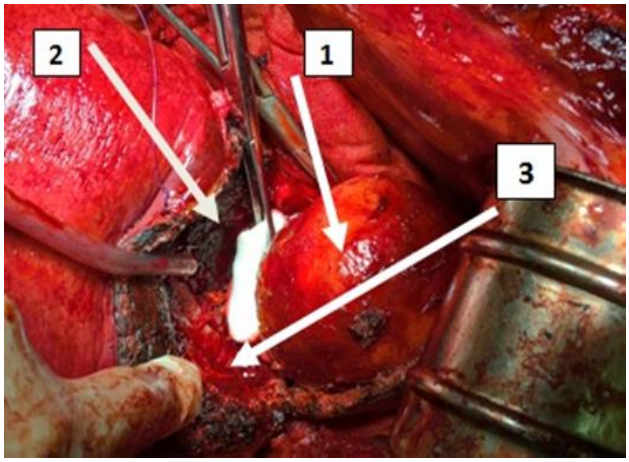

Figure 6: Mobilization of the 7th segment of liver with a cyst from the right hepatic vein.
1-Echinococcal cyst, 2-cyst bed and 3-right hepatic vein.

The bed of the resected cyst with 7th segment of the liver is coagulated (Figure.7). Control for hemostasis, bile discharge was performed.

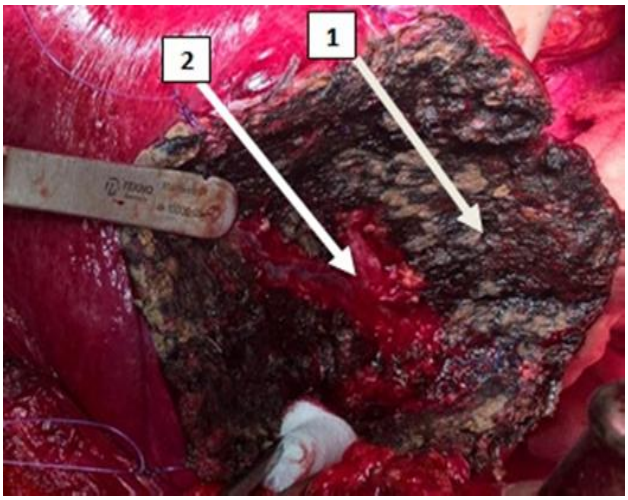

Figure 7: The bed of the removed echinococcal cyst from 7th segment of the right lobe of the liver.
1-The bed of an echinococcal cyst and 2-right hepatic vein with branches to 6 and 7 segments.

Drains are installed in the subdiaphragmatic space and the subhepatic space on the right. The layered stitches on the wound. Cleaned with antiseptic and placed an aseptic dressing over the wound.

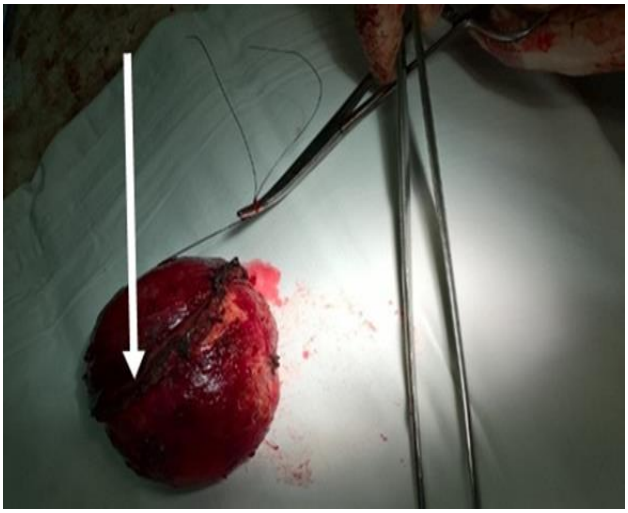

Figure 8: The removed echinococcal cyst from the 7th segment of the liver is indicated by an arrow.

DISCUSSION

The classification of liver cysts as either simple or complex is crucial for guiding treatment decisions. Simple cysts are characterized by their thin, smooth walls and cuboidal epithelial lining, producing a bile-like fluid. This category encompasses several conditions, including congenital cysts, biliary hamartomas, Caroli disease and polycystic liver disease. In contrast, hydatid or echinococcal cysts represent a complex parasitic type of liver cyst, resulting from infection with the *Echinococcus granulosus* parasite.[11]

Even though, echinococcosis is considered as a rare disease in some regions, yet it poses a significant challenge due to its high potential rate for morbidity and mortality as it involves the critical hepatic vasculature. The distribution of hepatic cysts across different liver segments influences the risk profile associated with each location. Especially, segments VII and VIII of the right lobe show the highest concentration of cysts (46%), presenting a twofold risk: firstly, their proximity to the diaphragm may necessitate a transthoracic surgical approach and increase the risk of hepato-thoracic injury; secondly, these cysts can affect the caval-suprahepatic venous system.[12]

Treatment for CE involves four main approaches: i) surgical resection, ii) Percutaneous Aspiration, Injection, and Re-aspiration (PAIR), iii) medical therapy using anthelmintic drugs (such as albendazole or mebendazole), and iv) a conservative "wait-and-see" strategy for asymptomatic, inactive cysts. Studies suggest that pre-operative anthelmintic treatment improves outcomes in patients undergoing surgery for CE (CE). Currently, albendazole is the first-line medical therapy. Furthermore, research indicates that combining albendazole with praziquantel enhances parasite killing and improves cure rates, reducing recurrence compared to albendazole monotherapy.[13]

Retrospective studies indicate that surgery, often combined with chemotherapy, remains the most common treatment approach for CE.[14]

Echinococcal cyst surgery aims to eliminate the parasite, remove the cyst, destroy the germinal layer, and seal the remaining cavity. Regardless of the surgical method, meticulous wound packing is crucial, employing scolicidal solutions (e.g., hypertonic saline, povidone-iodine, or albendazole) to prevent parasite spreading. Major surgical risks include postoperative hemorrhage, bile leakage, fistula formation, infection (ranging from local wound infection to sepsis), pulmonary complications (pneumonia, embolism), anesthetic complications, and even death.[15]

The decision to proceed with surgical intervention in these cases was based on, world health organization guidelines on Surgical treatment of CE.[16] The successful outcomes of the TVI used in treatment of our two patients who had echinococcal liver cysts, underscore the importance of a multidisciplinary approach and innovative surgical techniques while addressing complex hepatobiliary pathologies.

TVI, a technique which is refined over decades of surgical practice, is employed in these 2 cases, isolating the liver from the circulation, which merely aims at

preventing major complications like abundant bleeding and air embolism while minimizing the intraoperative blood loss and ultimately optimising surgical outcomes. It is done by temporarily occluding the hepatic inflow and outflow by applying safety vascular ligatures on the supra hepatic part, sub hepatic part of the inferior vena cava and on the hepatoduodenal ligament as well.

Both patients presented with hydatid cysts in close proximity to the right hepatic vein, located in segment VII. This location aligns with findings from numerous studies that indicate a higher prevalence of hepatic cysts, including hydatid cysts, in the right lobe of the liver, particularly segments VII and VIII.[17,18] These studies often attribute this distribution to the anatomical characteristics of the liver and the flow of the portal venous system. And in our case, both patients had focal formations intimately adjacent to large liver vessels and we were able to successfully facilitate meticulous dissection which allowed precise delineation of resection margins and facilitate safe manipulation of cysts which are intimately associated with major hepatic vessels. Advanced imaging modalities including CT, MRI and ultrasound played a pivotal role in pre-operative assessment and surgical planning. By these, accurate localization of the cystic lesions within the 6 and 7 liver segments were found as well as assessment of their proximity to major hepatic vasculature. Most importantly, the above-mentioned imaging studies along with intraoperative decision making and ensured optimal outcomes in both the patients.

The duration of the operation in both cases was about 6 hours. Intraoperative blood loss averaged about 400 ml. This level of blood loss is considered to be low, especially when compared to studies involving major hepatic resections.[19] This finding emphasizes the effectiveness of TVI in minimizing blood loss during complex liver surgery. For example, Cherqui et al reported mean blood loss of 900 ml in major resections; but those were for malignant tumors; and may not be comparable. Despite the presence of "linear traction" of the inferior vena cava in one of the patients, thanks to the use of TVI, it was possible to minimize intraoperative blood loss and stop bleeding by applying a vascular suture. TVI provides quick and reliable control over blood loss, which in turn allowed for a safe and more aggressive approach to the removal of focal formations intimately adjacent to large liver vessels. TVI completely turns off the liver from circulation and prevents the possibility of bleeding but may be accompanied by hemodynamic disorders, including decreased cardiac index and increased peripheral vascular resistance.[20] However, in these cases, the duration of liver ischemia was limited to 15 minutes, which likely contributed to the hemodynamic stability observed during the procedures. Such short periods have not been associated with worse outcomes.[21] This is likely also due to the overall good health of both patients before surgery. Usually, a hemodynamic response leads to a decrease in the heart index by 40% and an increase in peripheral vascular resistance by 80%. In our case, the duration of turning off the liver from blood circulation was up to 15 minutes when suturing the place of "linear traction" of the inferior vena cava. Hemodynamically, the operations were stable. There were no liver disfunction and other complications in both patients in the postoperative period. Both patients were discharged from the surgical hospital in a satisfactory condition for 10 and 15 days respectively in the postoperative period. This is comparable with outcomes shown in the lack of major morbidity or mortality further supports the safety and efficacy of the surgical approach employed.[22,23] During the pathohistological examination of macro-preparation, the diagnosis of echinococcal cyst was confirmed in both cases which aligns to a very high certainty with MRI and ultrasound imaging used. There were no relapses of the disease, as well as repeated admissions to the hospital with a follow-up period of up to 1.5 years. Other studies have shown that recurrence rates are relatively low, but longer follow-up periods are relatively low, but longer follow-up periods are needed.[24] Although there was no recurrence in our case series, 1.5 years is not enough for the certainty's.

CONCLUSION

A segmental resection of the liver for an echinococcal cyst using TVI was performed. It is a game-changer in the surgical management of CE, allowing a safe and highly efficacious approach to remove the cyst.

This procedure involves clamping the right triangular and coronary ligaments of the liver and mobilizing inferior vena cava in the supra and subhepatic sections as well as the hepatoduodenal ligament using a thread. Then subsequent mobilization of 6 and 7 segment of liver from inferior vena cava with lower hepatic veins was conducted. Later trans-section of liver parenchyma, bipolar and monopolar coagulation and precision stitching of vascular and biliary structures was carried out. The most difficult part of this procedure was to separate the cyst from the right hepatic vein and its branches to 6 and 7 segment which could easily rupture during surgery due to its complex vasculature. But we were supposed to done it successfully with a minimal intraoperative blood loss of 400 ml in both the cases and without post operative complications using this method. Finally, stitches were made on the wound after installing drains in the sub-diaphragmatic and subhepatic space on the right side. However, this technique requires a high degree of surgical skill and expertise, as misapplication of the procedure may result in such potential consequences as hemodynamic instability and vascular injury. Additionally, the procedure may be difficult to perform when the patient presents with tissue conditions or anatomical abnormalities, necessitating careful patient selection and extensive preoperative evaluation. Nevertheless, there can be no doubts about the advantages of TVI in treating CE. The method's power to

ensure rapid and consistent control over the hepatic circulation allows prevents surgeons from approaching the complex vascular in uncertain terms and confidently perform complete excision without compromising the liver function. In the future, the incorporation of TVI will contribute to a new standard of care for cystic echinococcal patients. This method is a safe, efficient, and minimally invasive way to perform surgical treatment in this pathology.

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