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CASE REPORT

Ex vivo Liver Autotransplantation for Alveolar Echinococcosis with Brain and Lung Metastases: A Case Report

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Abstract: Ex vivo liver resection and autotransplantation (ELRA) in extensive abdominal alveolar echinococcosis (AE) is a relatively rare treatment. We present the first case, ever reported in Kazakhstan, of successfully performed ELRA of a patient with extensive abdominal AE with invasion into the inferior vena cava (IVC). A 64-year-old woman was diagnosed with AE of the brain, alveococcectomy from the left frontal lobe was performed. At the same time, liver AE metastased to the lungs and brain was diagnosed. The patient was admitted to our center for the following surgical treatments: (1) laparotomy, (2) extended left hemihepatectomy with resection of IVC ex vivo, (3) plastic surgery of IVC with a synthetic prosthesis, (4) orthotopic transplantation of the remnant right lobe of the liver. The postoperative period proceeded smoothly. The patient was mechanically ventilated for 1.5 days and prescribed strict bed rest with in-bed activity for up to 11 days. Empirical antibiotic therapy was initiated as follows: meropenem 1000 mg three times a day for 10 days, omeprazole 40 mg once a day for 14 days, anticoagulant therapy was not administered in the first post-operation day due to high risk of bleeding. Clexane was prescribed subcutaneously once a day: 0.2 mL for 2 days, then 0.4 mL for next 10 days. Daily transfusion of 100 mL of 10% Albumin was performed to prevent hypoalbuminemia. Drainage systems were removed on the 9th day. On the 12th day, the patient was transferred from the intensive care unit to a hospital room, and discharged with improvement on the 20th day. During a 6-month follow-up, no relapse was observed. Radical surgery for widespread AE, in this case – ELRA, is the preferred treatment, since it does not require immunosuppressive therapy and is quite feasible even with invasion of the portal vein bifurcation, IVC and hepatocaval confluence.

Keywords: alveolar echinococcosis, inferior vena cava resection, liver autotransplantation

Introduction

Kazakhstan is a highly endemic region for alveolar echinococcosis (AE): according to data for recent decades, 800–1000 cases per year are noted annually.[1] The treatment of patients with parasitic liver diseases is constantly discussed in the literature, but today there are no clearly established surgical standards.[2] Some authors assess this variant of AE as "parasitic liver cancer" due to infiltrative growth, the possibility of metastasis, as well as a high recurrence rate after surgery.[2–4] Surgical treatment of extensive liver AE with the spread of the pathological process to borderline or distant vital organs is particularly difficult. The purpose of our case report was to disseminate our clinical experience of successfully performed liver resection and autotransplantation (ELRA) in a patient with extensive abdominal AE invaded into the inferior vena cava (IVC) – first ever reported in Kazakhstan.

Case Presentation

Materials and Methods

A 64-year-old woman of Kazakh nationality was diagnosed with AE of the brain for the first time in April 2021, alveococcectomy from the left frontal lobe was performed in the neurosurgery department. At the same time, liver AE

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with metastases to the lungs and brain was diagnosed. During the period between 2021 and 2023, the patients received several courses of Albendazole, with the dose of 400 mg twice a day. In July 2023, the patient was admitted to the Department of General and Thoracic Surgery of JSC "National Scientific Medical Center" (Astana, Kazakhstan) with extensive liver AE metastased to the lungs and brain (P4NxM1). The ELRA in extensive abdominal AE was implemented in the Department. According to the patient's history, the patient was a permanent city occupant, not having dogs and cats during the last decades, and not occupied in any farm-related job or activity.

Diagnostic Assessment and Surgery

Contrasted computed tomography of the abdominal cavity and retroperitoneal space was performed. The computed tomography scans revealed cyst-like area (of parasitic genesis) of the left lobe of the liver (Figure 1A and B), cysts of the left kidney of category I according to Bosniak,[5] chronic cholecystitis, chronic pancreatitis, and osteochondrosis of the lumbar spine. Computed tomography scans of the chest and mediastinal organs revealed structures in the lower lobes of both lungs (probably parasitic aetiology, abscess is not excluded), chronic bronchitis, and osteochondrosis of the thoracic spine. The patient underwent the following surgical treatments according to the planned scheme: 1) laparotomy, 2) extended left-sided hemihepatectomy with ex vivo resection of the IVC, 3) plastic surgery of the IVC with a synthetic prosthesis, 4) orthotopic transplantation of the dominant right lobe of the liver.

The surgical treatment was performed by two teams of surgeons: the first team made the laparotomy to provide an operational access and the second team performed the main stages of the surgery. Intraoperative access was performed by Mercedes type laparotomy. During the revision of the abdominal cavity, a slight adhesive process was revealed and acutely dissected, there was no effusion. The liver was not enlarged, and the entire anatomical left lobe of the liver was occupied by an echinococcal cyst with spread to the segments SIV, SVIII, with signs of invasion along the anterior semicircle of the IVC, the middle and left hepatic veins in the area of confluence with the IVC (confirmed by computed angiography in the preoperative period). A metastatic lesion along the diaphragm surface of the peritoneum, up to 0.8 cm in size, was excised. The lymph nodes were not affected by the lesions and remained intact. The rest of the abdominal organs was without abnormalities. Next, the second team performed ex vivo complete resection of liver after applying vascular clamps to the IVC along the planned boundaries, as well as to the elements of the hepatoduodenal ligament, and crossing the vessels. The first team then repaired the IVC defect with a vascular prosthesis D-20mL, with temporary restoration of blood flow from the portal vein system to the caval system. The second team washed the vessels of the removed liver with a total of 4 L of Custodiol (Franz Kohler Chemie GmbH, Germany) to elevate tissue resistance to the hypoxia. The final ex vivo liver resection was performed: the demarcation line between the left and right lobes was

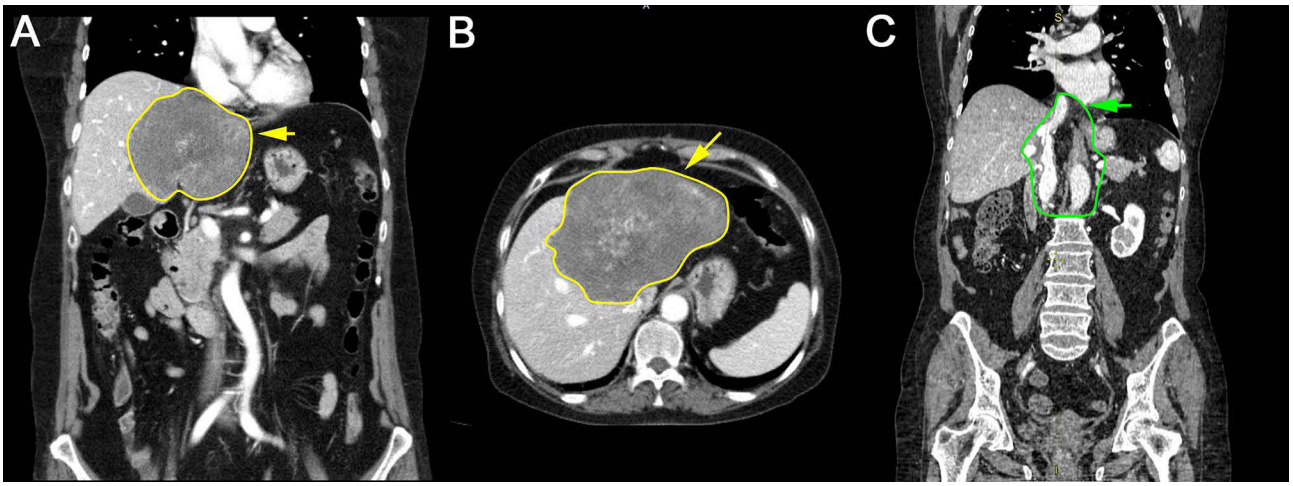

Figure 1 Computed tomography of the abdominal organs and retroperitoneal space with contrasting. (**A** and **B**) The image of cyst of the left lobe of the liver (of parasitic origin), dimensions are 19×15 cm with uneven clear contours. (**A**) Frontal plane, (**B**) Axial plane, yellow contours and arrows indicate the cyst margins. (**C**) The image in dynamics three months after the surgery, frontal plane, no relapse as shown by bright green contour and arrow.

determined (including the distance of 1–2 cm away from the edge of lesions), the dominant right lobe was cut along the line (final weight 560 g), and the left lobe was completely removed. Vessels of the right lobe were identified, washed with 400 mL of 10% Albumin solution (Baxter, Austria) to maintain oncotic pressure and cell integrity, and then orthotopically inserted into the abdominal cavity. Next, consecutive single-row continuous vascular anastomoses with atraumatic sutures (all from Johnson & Johnson, USA) were performed: hepatic vein (Prolene 4.0), portal vein (Prolene 5.0), hepatic artery (Prolene 9.0), common bile duct (Monocryl 5.0). At the same time, the IVC was restored by continuous suturing. Doppler ultrasound of the liver vessels was performed intraoperatively, and the blood supply through the portal vein and hepatic artery system was satisfactory. Intraoperative blood loss was 2000 mL. The total duration of the preliminary steps and the main surgical procedure was 685 min (~11.5 hours). The surgical mobilizing of the organ within the procedure lasted 4 hours 20 minutes, the prosthetic repairing of the IVC defect lasted 20 minutes, and the final resection of the right lobe of the liver and its orthotopic transplantation lasted 30 minutes.

Pathohistological conclusions were as follows: local hyalinosis with foci of dystrophic calcification, AE of the liver with infiltrative spread and multiple seeding foci of between nerve fascicles, with growth to the intrahepatic bile duct and arterial vessels. Productive granulomatous inflammation with Langhans giant cells. There were no signs of invasive spread of helminthiasis along the far edge of the resection.

The patient was on mechanical ventilation for 1.5 days as part of the therapeutic regimen. Extubation was performed as scheduled. To prevent transposition of the remnant liver lobe, the patient was prescribed bed rest while with in-bed activity for up to 11 days. In the postoperative period, empirical antibiotic therapy was initiated as follows: meropenem 1000 mg three times a day for 10 days, gastroprotective therapy by omeprazole 40 mg once a day for 14 days. Anticoagulant therapy was not administered on the first day post-operation due to a high risk of bleeding. Clexane (Sanofi-Aventis, France) was prescribed to prevent thrombosis of deep veins, subcutaneously at a dose of 0.2 mL once a day for 2 days then at a dose of 0.4 mL once a day for next 10 days. Daily transfusion of 100 mL of 10% Albumin was performed to prevent hypoalbuminemia. Drainage systems were removed on the 9th day.

Results

The postoperative period proceeded smoothly. On the 12th day, the patient was transferred from the intensive care unit to the ward, and then discharged with improvement on the 20th post-operative day. The patient was prescribed Albendazole after discharge. At present, we are on the 6th month of outpatient observation of the patient. According to abdominal computed tomography data, no recurrence is observed (Figure 1C) while the patient has refused operative treatment for metastatic lesions observed in the lung. Overall, the patient confirmed her agreement with the treatment plan before the surgery, was satisfied with all the treatments, had no complications, and did not express any complaints with regard to the treatment-related financial costs.

Discussion

While non-surgical approaches like focused ultrasound have already been described to treat non-resectable liver alveolar echinococcosis (AE),[6,7] it is often that the only way to treat such patients is the ex vivo liver resection and autotransplantation (ELRA).[8–10] However, there is relatively little experience in the world about ELRA in non-resectable AE. This is in part due to the endemic nature of the disease – Echinococcus multilocularis is extremely rare in many countries with a developed liver transplantation program.[3] On the other hand, the most frequently reported AE cases include regions and countries such as France, Germany, Poland, as well as China and Turkey.[11–13] Moreover, Asian countries show the highest rate of incidence and fatalities due to the disease.[14] Accordingly, significant efforts have been made by the Asian surgeons, especially Chinese, to improve the clinical outcome of the ELRA.[15]

The applicability of the treatment has been demonstrated in several studies. For example, it was performed with a good success rate in two Chinese hospitals, as reported in:[16] only in two out of six patients the in-hospital complications were observed, with no in-hospital lethality as well as without lethality after median 18.6 months follow-up. In another Chinese case series with 15 end-stage hepatic AE patients, the mortality rate was 13.3% (one fatality on day 12 after surgery and another death during 6-month follow-up period).[17] Comparable mortality rates were obtained for a much larger cohort of ELRA-subjected liver AE patients (n = 114): 7% for 30-days mortality and

13% for 90-day mortality.[18] Another Chinese case series reported 11 successful surgeries without in-hospital mortality; the median liver autograft weight and in-hospital stay were comparable with our case (690 vs 560 g and 15 vs 20 days, respectively).[19] Successful and uneventful surgical outcome were reported in the case of giant hepatocellular carcinoma, where the patient was subjected to the ELRA.[9] Similarly, in the end-stage hepatic AE with invasion into portal vein, the ELRA was performed with no adverse events observed during 3-year follow-up; however, in this case, the substantial portal vein reconstruction was needed intraoperatively.[8] Even in the case of advanced multi-organ AE lesions (eg affecting liver, lungs, heart), the ELRA may be a life-saving option despite a number of complications, as was reported recently.[10]

It is important to highlight that the overall success of the ELRA depends on whether all affected tissue has been removed prior to the liver autotransplantation. In cases of incomplete removal of the lesions, the recurrence is an actual threat.[4] Also, possible post-operative complications must always be carefully considered prior to the ELRA. For example, recently reported studies indicate that more attention must be given to possible vascular complications.[12] In patients with end-stage hepatic AE and severely compromised hepatocaval confluences, the complications include IVC stenosis and thrombus.[18] On the other hand, technical difficulties of the operation due to parasitic invasion of the main vessels, IVC, right atrium, and neighboring structures and organs limit the possibilities of ELRA.[4] Also, the post-operative observation may include recently proposed approaches like proteomic analysis for certain serum proteins (which have been found to be potential biomarkers for complications) in patients previously subjected to the ELRA.[20] Finally, the lack of procedural standards for liver resection/autotransplantation in hepatic cancer and cancer-like pathological states limits the success of the application of this approach.[2]

In addition, ethical issues are important when considering the radical surgery as the only available option. However, this aspect is managed by both the surgeons and the patient, and the final decision is also dependent on the possible (predicted) outcomes. Both uneventful (eg[10]) and eventful (eg[4]) cases post-ELRA may occur, and the risk should be assessed appropriately.

Our results highlight the need and feasibility of ELRA for non-resectable liver AE, regardless of the presence of invasion to the main vessels, porta hepatis, inferior vena cava, right atrium, and neighboring structures and organs. In many countries of low- and mid-development level, such type of surgery is rarely performed. For example, in Kazakhstan, which is the upper middle-income country, the presented ELRA case is the first successful clinical case ever reported. We suggest considering the radical surgery for such similar cases in Kazakhstan, taking into account that it is endemic for AE, and assuming careful diagnosis and pre-treatment evaluation of patients for possible post-operative complications have been done.

Conclusions

In conclusion, ex vivo liver resection and autotransplantation for extensive hepatic alveolar echinococcosis, even in cases of invasion into the bifurcation of the portal vein, inferior vena cava, and hepatocaval confluence is a reliable and preferable therapeutic strategy feasible in specialized clinical settings.

Data Sharing Statement

All data generated or analysed during this study are included in this published article.

Ethics Approval and Consent to Participate

This study has received ethical approval from a local ethical board of the National Scientific Medical Center. The institutional consent form for participation in publication was approved by the local ethical board. All methods and treatments were performed in accordance with the relevant guidelines and regulations.

Consent for Publication

Written informed consent was obtained from the patient. Assent from minors was not obtained because it is not applicable.

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Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

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Disclosure

The authors report no conflicts of interest in this work.

References

1. Abdybekova A, Sultanov A, Karatayev B, et al. Epidemiology of echinococcosis in Kazakhstan: an update. *J Helminthol*. 2015;89(6):647–650. doi:10.1017/S0022149X15000425
2. Yang X, Lu L, Zhu WW, et al. Ex vivo liver resection and auto-transplantation as an alternative for the treatment of liver malignancies: progress and challenges. *Hepatobiliary Pancreat Dis Int*. 2024;23(2):117–122. doi:10.1016/j.hbpd.2023.10.007
3. Barth TFE, Casulli A. Morphological characteristics of alveolar and cystic echinococcosis lesions in human liver and bone. *Pathogens*. 2021;10 (10):1326. doi:10.3390/pathogens10101326
4. Pielok Ł, Karczewski M, Cierach W, Zmora P, Lenartowicz E, Stefaniak J. Portal hypertension as a result of the incomplete surgically treated advanced alveolar echinococcosis: a case description. *BMC Gastroenterol*. 2020;20(1):176. doi:10.1186/s12876-020-01320-0
5. Muglia VF, Westphalen AC. Bosniak classification for complex renal cysts: history and critical analysis. *Radiol Bras*. 2014;47(6):368–373. doi:10.1590/0100-3984.2013.1797
6. Imankulov SB, Fedotovskikh GV, Zhampeissov NK, Yerlan M, Shaimardanova GM, Knaub LV. Treatment of liver alveococcosis with high-intensity focused ultrasound. *Ultrason Sonochem*. 2015;27:707–711. doi:10.1016/j.ultsonch.2015.05.022
7. Zhampeissov N, Manap E, Rustemova K, et al. High-intensity focused ultrasound ablation: a non-surgical approach to treat advanced and complicated liver alveococcosis. *J Med Ultrason*. 2019;46(2):251–255. doi:10.1007/s10396-018-0914-x
8. Yilihaer Y, Wang M, Aji T, Shao Y, Aikebaer A, Mahmood A. Successful interventional therapy for portal vein stenosis after ex vivo liver resection and autotransplantation in end-stage hepatic alveolar echinococcosis with cavernous transformation. *Ann Transplant*. 2024;29:e944851. doi:10.12659/AOT.944851
9. Kanwal N, Haider J, Haider S, Iqbal M, Amir K, Dar FS. EX vivo liver resection and auto transplantation for a giant hepatocellular carcinoma: a case report. *Int J Surg Case Rep*. 2024;125:110573. doi:10.1016/j.ijscr.2024.110573
10. Ruze R, Jiang T, Zhang W, et al. Liver autotransplantation and atrial reconstruction on a patient with multiorgan alveolar echinococcosis: a case report. *BMC Infect Dis*. 2024;24(1):659. Erratum in: *BMC Infect Dis*. 2024;24(1):790. doi:10.1186/s12879-024-09690-6. doi:10.1186/s12879-024-09545-0
11. Baumann S, Shi R, Liu W, et al. Worldwide literature on epidemiology of human alveolar echinococcosis: a systematic review of research published in the twenty-first century. *Infection*. 2019;47(5):703–727. Erratum in: *Infection*. 2022;50(1):287-288. doi:10.1007/s15010-021-01695-6. doi:10.1007/s15010-019-01325-2
12. Mamuti A, Tulading A, Zhang YF, et al. Insight into the history and trends of liver transplantation for echinococcosis: a bibliometric analysis. *World J Hepatol*. 2024;16(12):1493–1504. doi:10.4254/wjh.v16.i12.1493
13. Akyuz M, Avcioglu H. Evaluation of the prevalence of Echinococcus multilocularis in free-roaming dogs in the human alveolar echinococcosis endemic region of Turkey. *Acta Trop*. 2025;107560. doi:10.1016/j.actatropica.2025.107560
14. Deplazes P, Rinaldi L, Alvarez Rojas CA, et al. Global distribution of alveolar and cystic echinococcosis. *Adv Parasitol*. 2017;95:315–493. doi:10.1016/bs.apar.2016.11.001
15. Aini A, Lu Q, Wen H, et al. Particular Chinese contributions to extracorporeal liver surgery. *Hepatobiliary Pancreat Dis Int*. 2025;24(1):57–66. doi:10.1016/j.hbpd.2024.12.005
16. Lv T, Xu G, Xu X, et al. A novel remnant liver-first strategy for liver autotransplantation in patients with end-stage hepatic alveolar echinococcosis: a retrospective case series. *Int J Surg*. 2023;109(11):3262–3272. doi:10.1097/JS9.0000000000000604
17. Wen H, Dong JH, Zhang JH, et al. Ex vivo liver resection and autotransplantation for end-stage alveolar echinococcosis: a case series. *Am J Transplant*. 2016;16(2):615–624. doi:10.1111/ajt.13465
18. Maimaitinijiati Y, AJi T, Jiang TM, et al. Approaches to reconstruction of inferior vena cava by ex vivo liver resection and autotransplantation in 114 patients with hepatic alveolar echinococcosis. *World J Gastroenterol*. 2022;28(31):4351–4362. doi:10.3748/wjg.v28.i31.4351
19. Wang C, Qiu Y, Wang W. Application of ex vivo liver resection and autotransplantation in treating Budd-Chiari syndrome secondary to end-stage hepatic alveolar echinococcosis: a case series. *Medicine*. 2021;100(34):e27075. doi:10.1097/MD.0000000000027075
20. Sadjjadi FS, Ahmadi N, Rezaie-Tavirani M, Zali H. Following up of surgical treated human liver cystic echinococcosis: a proteomics approach. *Iran J Parasitol*. 2021;16(1):11–22. doi:10.18502/ijpa.v16i1.5507