COMBINED SURGERY OF LUNG ECHINOCOCCOSIS

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Abstract

Background. Pulmonary cystic echinococosis, a parasitic disease, is a health care problem in developing countries. In this study, we evaluated outcomes of patients with pulmonary hydatid disease who were treated in our department.Study was performed to compare results of surgical treatment and complications of patients with unilateral or bilateral thoracic and combined pulmonary cystic echinococosis.

Methods. This cross-sectional analysis of a prospective study was conducted in the Department of Thoracic and Pediatric Surgery, Scientific Center of Surgery, Almaty, Kazakhstan among 598 patients with pulmonary cystic echinococosis, who had surgical treatment with various surgical methods, depending on the prevalence of echinococcosis, as follows: right lung in 357 (59.5%) patients, left lung in 243 (40.5%) patients, bilateral in 95 (15.8%) patients, and complicated echinococcosis in 317 (52.8%) patients. Length of stay per hospital stay has been decreased (p 0.0001) by video-thoracoscopicechinococcectomy with the high-energy laser treatment of cyst, than after echinococcectomy by cyst treatment with povidone-iodine. Treatment with formalin presented the longest hospital stay (p 0.0001).

Results. Comparative analysis of patients with uncomplicated and complicated pulmonary cystic echinococosis showed a high frequency of postoperative complications associated with complicated echinococcosis (OR = 2.2, p 0.0001).

Conclusion. Despite the success of surgical treatment of pulmonary cystic echinococosis, issues of intraoperative dissemination and safety remain, and treatment success rates can be improved. These factors require further prospective multicenter studies.

Introduction

Pulmonary cystic echinococcosis (PCE) has no clinicalpresentation and may cause impassable cough, coloredsputum, hemoptysis, and fever.[1,2] X-ray and Computed tomography (CT) can detectlung abscess, bronchoscopy can detect cystic lesion,and serological testing can detect antibody titer to Echinococcus granulosus.[2-5] Cystic echinococcosis isthe most common type, and represents 95% of thecases. Estimated cases worldwide was 2 to 6 million, and the mortality rate was 2% to 4% per 100.000 worldpopulation.[6]

Endemic cystic echinococcosis primarilyoccurs in Mediterranean countries, Central Asia, North and East Africa, Australia, and South America.[6,7] Despite of development of new oral anti-parasiticmedicines; only surgical approaches were able to proveits effectiveness in the treatment of cystic echinococcosis. Other important challenges that deserve tostudy are postoperative complications and recurrentechinococcosis.[8,9] The purpose of this study is to explorethe number of postoperative complications associatedwith various surgical treatments of complicated anduncomplicated PCE.

Materials and methods

This cross-sectional analysis of aprospective study was conducted in the Departmentof Thoracic and Pediatric Surgery, Syzganov National Scientific Center of Surgery, Almaty, Kazakhstan from 2018 to 2024.Ultrasound (US) or CT and other complete data availableobtained from the patients with primary PCEwere included in this review. Adult age≥18 and <70 years old were also included. Patientsreferred from any out-patients department or hospitals throughout Kazakhstan were also included. Presenceof hydatid cysts, any size on the US or CT, were the indica-

tions for echinococectomy.

Exclusion criteriawere pregnant women, patients with fever and activepulmonary tuberculosis, HIV positive with HIVsymptoms, as well as with primary or secondary lungor liver cancer.

We analyzed treatment results of 598 patients withPCE who were hospitalized in thedepartment of thoracic surgery. Two hundred eightythree patients had uncomplicated echinococcosis, and317 patients had complicated echinococcosis, due tofestering echinococcosis cysts in the bronchus.Patients ranged in age from 32 to 67 years andincluded 74 (12.4%) women and 524 (87.6%) men. Data were collected prospectively from the InstitutionalEchinococcosis Registry.

Ethical approval. The study protocol was approved by our Institutional Local Research EthicsCommittee (2023), and the study protocol wasdeveloped to conform to the ethical standards of theDeclaration of Helsinki. We received informed consentfrom all participants in the study.

Statistical Analysis. Were conducted with SPSS software version18.0. Z-statistic for analysis of maincharacteristics surgical patients, complications anddeaths, Chi-square test for analysis of surgery methods,the odds ratio (OR) for analysis of complications anddeaths. A p<0.05was used to determine significance. Continuous data(hospital stay, days) are presented as mean standarddeviation (SD) or median and categorical data arepresented as frequency in percentage. Comparisons ofpatients' characteristics and outcomes were conductedin the 2 patient groups with uncomplicated andcomplicated cases of PCE.

Results

Combined lesions onlungs and liver was seen in 136 (22.7%) patients, lungsand other organs in 40 (6.7%) patients, 22 (3.7%) ofthem in the lesser sac, 8 (1.3%) of them in the spleen,6 (1%) of them in the abdomen, and 4 (0.7%) in the greater omentum (Table 1). Unilateral common lesion ofthe lung with echinococcosis is statistically important,followed by bilateral lesion (p≤0.0001), and combinedlesion of the lung, liver, and other abdominal organs,(p≤0.0001).

Table 1.
Main features

	Unilateral lesion of the lungs		Bilateral lesion of the lungs		Combined lesion of lungs, liver and etc.		Chi-squared	P value
	n	%	n	%	n	%		
Number of patients	327[a,c]	54.7	95 [a,b]	15.9	176 [b,c]	29.4	44.5* 6.0 29.4*	0.0001 - 0.0001
Right lung	195	59.6	-	-	88	14.7	49.1*	0.0001
Left lung	132	40.4	-	-	32	5.3	14.1*	0.0002
Both lungs	-	-	-	-	12	2	-	-

*Statistical significant difference p≤0.05. a - comparison between patients with unilateral and bilateral involvement; b- bilateral involvement and combined lungs and extrathoracic involvement; c - unilateral involvement and combined lungs and extrathoracic involvement

Operative procedures. Organ-preserving surgerywas performed, which is more statistically significant(p≤0.0001) than frequency of lung resection (Table 2).

Table 2.
Organ-preserving surgery consisted of closed echinococcectomy by methods Delbe's, echinococcectomy or lung resection by Bobrov Spasocucotsky's or Vishnevsky's methods

	Organ-preserving surgery		Lung resection		CI	Chi-squared
	n	%	n	%		
Methods of surgery	536	89.6	62	10.4	[68.4;85.9]	229.7*
By Delbe	281	47	-	-	-	-
By Bobrov-Spasocucotskii	11	1.8	37	6.2	[17.6; 25.6]	0.3

by Vishnevskii	7	1.0	25	4.2	[17.6; 25.6]	0.3
One stage bilateral thoracoto-my with echinococcectomy	23	3.8	-	-	-	-
One stage bilateral video-assisted thorascopicechinococcectomy	30	5	-	-	-	-
One stage bilateral thoracotomy with echinococcectomy with trans medistinal access	9	1.5	-	-	-	-
Two stage bilateral thoracotomy with echinococcectomy	33	5.5	-	-	-	-
One stage thoracotomy with laparotomy	59	9.9	-	-	-	-
Two stage thoracotomy with laparotomy	84	14.1	-	-	-	-
Fillingacyst						
By Kulakeev	176	2.3	-	-	-	-
With capitonage	43	7.2	-	-	-	-
the treatment of fibrous cyst capsule cavity						
High-energylaserbeam	109	18.2	-	-	-	-
Low-frequencyultrasound	97	16.2	-	-	-	-
formalin	180	30.1	-	-	-	-
Povidone-iodine	200	33.4	-	-	-	-
* Statisticalsignificantdifferencep≤0.05						

Lung resection was carried out in 23 (3.8%) casesof festering cysts; in 18 (3%) cases of echinococcosisoccupying the volume of almost the entire fraction(2 lobes) with irreversible coarse peri focal changes and fibrosis in the surrounding pulmonary tissue; andin 13 (2.2%) cases of echinococcosis with excessivemultiplicity lesion of one or 2 lbes. Organ-preserving surgery consisted of Delbe closed echinococcectomy, echinococcectomy or lung resection by Bobrov-Spasocucotsky's or Vishnevsky's methods. (Figure1.)

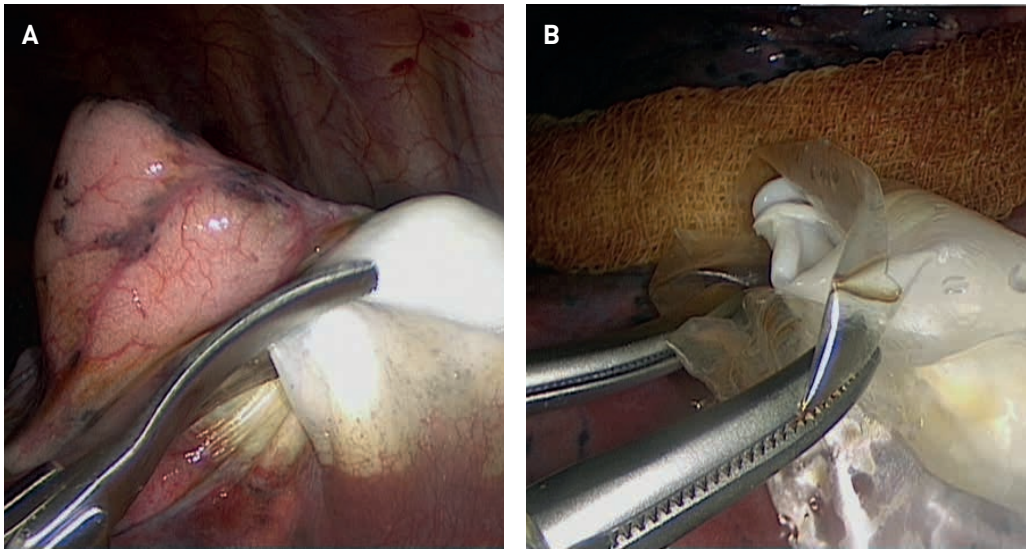
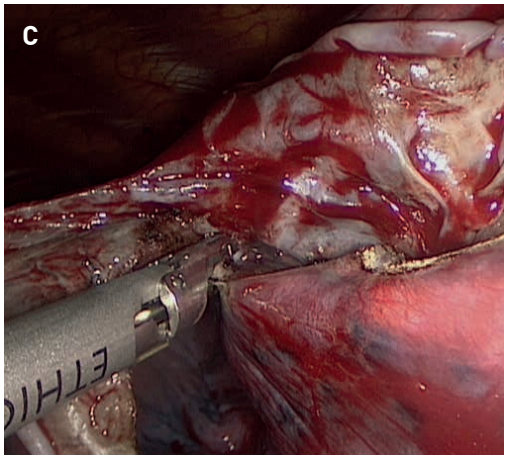
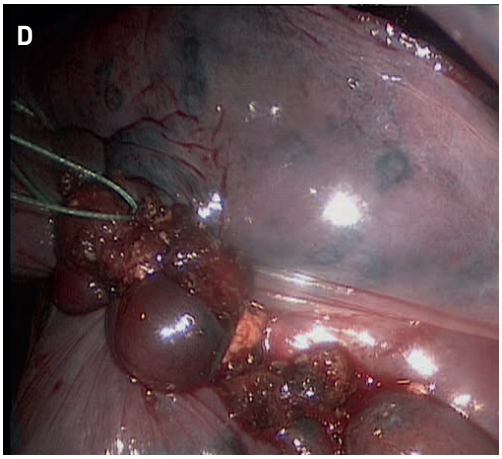


Figure 1.
Video-assisted thorascopicechinococcectomy
A. Opening of the echinococcal cyst fibrous capsule after puncture aspiration of the contents;
B- Removing chitin membrane;
C- Excision of the outer fibrous capsule after treatment with 10% povidone-iodine;
D-capitonage echinococcosis cyst bed.





The treatment of fibrous PCE cyst capsule cavity was performed using a high-energylaser beam or using low-frequency ultrasound. Theeffectiveness of these methods has been compared tothe result of anthelmintic treatmentwith formalin orpovidone-iodine. Filling a cyst of fibrous capsule with vertical half purse-string suture by Kulakeev's method was carried out in 176 cases, 50% of them with PCE complications. Capitonage was carried out through a combination of horizontal and vertical sutures in 21 patients with complicated cysts (12 suppuration, 4 chitin membrane detachments, 2 rupture of cyst into bronchus, one rupture of cyst into the pleural cavity, and 2 pulmonary hemorrhages).

In patients with bilateral echinococcosis adhered to tactics, 2-stage bilateral thoracotomy was performed with an interval of 3, 6, or 8 weeks between them, depending on the severity. In patients with right PCEand upper segment liver right lobe, simultaneous one-stage thoracotomy with diaphragmotomy and echinococcectomy of the lung and liver were performed. In 44 (7.4%) patients with bilateral lesion of the lungs and spleen and liver echinococcosis, after phased thoracotomy with lung echinococcectomy (with an interval of 1 to 2 months between them), next-stage laparotomy was carried out to excise echinococcosis cysts of the abdominal parenchymatous organs. Of these, 27 patients had one-stage surgery; the others had 2-stage surgery with an interval of 4 to 8 weeks between them. In 40 (6.7%) patients with combined PCE- and abdominal organs, after thoracotomy and lung echinococcectomy, they received second-stage laparotomy and echinococcectomy from the lesser sac in 22 (3.7%) patients, from the abdomen in 6 (1%) patients, from the greater omentum in 4 (0.7%) patients, and with a splenectomy in 8 (1.3%) patients. One-stage bilateral video-assisted thoracoscopic echinococcectomy was performed in 30 (5%) patients with PCE. Organ-preserving video-assisted thoracoscopic echinococcectomy is performed under general anesthesia with separate intubation of the bronchi, which allows the surgeon to shut down the lung in the vents on the operative side. The cyst is covered with povidone-iodine wet napkins to prevent inadvertent implantation of scolices or daughter cysts. The pipe tool punctures the cyst through a thoracoport with hydatidic fluid aspirates; without removing the needle, 10% solution of povidone-iodine as scolicidal agent injected (nearly the same amount of the fluid aspirated) for 3 minutes. The fibrous capsule is opened and the chitin membrane is removed. The fibrous capsule cavity is eliminated depending on the size of the application clips or suturing.

A method of removing bilateral echinococcosis cysts of the lungs through transmediastinal access, was developed by our center. In bilateral lung echinococcosis when hydatid cysts located in the upper lobe, and in any part of the other lung, we carry out a one-sided lateral thoracotomy, hydatid cyst removed from one lung, and then performed resection of retrosternal mediastinal pleura, cyst of the upper lobe of the other lung moved to retrosternal mediastinal approach and then performed echinococcectomy. Then 2 pleural cavities

drained by 2 drainage tubes, one tube in the pleural cavity on the side of the thoracotomy, and second drain tube going through mediastinal approach to other pleural cavity, outputting the end of the tube through the chest wall on the side of thoracotomy. The advantages of this method are that the one-stage bilateral echinococcectomy using transmediastinal approach reduces the cosmetic defect and reduces pain. This method was used in 9 (1.5%) patients, who had no postoperative complications.

Postoperative outcomes. A comparative study of the postoperative period features and the long-term results of treatment with a high-energy laser (HEL) were carried out in comparison with the treatment of cyst by formalin or povidone-iodine solutions (Table 3).

Table 3.
Comparative characteristic of the postoperative period

	A treatment method of fibrous capsule				
	HELB	**Povidone - iodine**	**formalin**	**Z statistic**	**P value**
Number of complications	12 (2.0%)[a, b]	44 (15.5%) [a, c]	94 (29.7%)[b, c]	1.5 4.1* 3.2	- 0.0417 -
Stay in hospital (bed-day)	5.5±0.8 [a, b]	16.3±2.0 [a, c]	19.4±2.1 [b, c]	18.2* 22.6* 8.2*	0.0001 0.0001 0.0001
* Statistical significant difference p≤*0.05*					

The worst results were after treatment of fibrouscapsule with formalin solution (p<0.0417). Thefrequency of complications after cyst treatment with HEL and povidone-iodine did not reach a statisticallysignificant difference (p≥0.05). The treatment methodfor echinococcosis cyst with 10% povidone-iodine isthe most simple, safe, and effective method. Presentlyin our center, HEL or a 3-time treatment with 10%povidone-iodine results in the cyst drying up; if apatient has an allergy to iodine, we treat the cyst with70% alcohol.

Length of stay per hospital stay has been decreased(p<0.0001) by video-thoracoscopic echinococcectomywith the HEL treatment of cyst, than after echinococcectomy by cyst treatment with povidoneiodine; finally, treatment with formalin presented thelongest hospital stay (p<0.0001).

Complications. We noted postoperative complications in 139 (23.2%) of 598 operated patients. Complications resulted in death in 4 (0.6%) patients. In 45 (16%) of 281 patients with uncomplicated PCE, postoperative complications occurred, including one death from cyst removal with formalin. Complications from PCEoccurred in 94 (29.7%) patients of 317 (53%); 3 of these resulted in death. Comparative analysis of patients with uncomplicated and complicated PCEshowed a high frequency of postoperative complications associated with complicated echinococcosis (OR = 2.2, p<0.0001). Mortality frequency has a direct relationship with complicated echinococcosis, but this relationship was not statistically significant (Table 4).

Table 4.
Surgical complication frequency

	The course of pulmonary cystic echinococcosis				
	Uncomplicated	**Complicated**	**OR**	**Z**	**P value**
Complications	45 (16.0%)	94 (29.7%)	2.2	3.9*	0.0001
Deaths	1 (0.2%)	3 (0.5%)	2.7	0.8	-
* Statistical significant difference p≤*0.05*					

Complications included reactive pleurisy in 47 (7.9%) patients, suppuration of the postoperative wound in 37 (6.2%) patients, bronchial fistula in 15 (2.5%) patients, and pneumonia in 14 (2.3%) patients. Less common complications were pleural empyema in 8 (1.3%) patients, residual cavities in the lung in 8 (1.3%) patients, pulmonary heart disease in 6 (1.0%) patients, and hemorrhage in 1 (0.2%) patient. (Figure 2)

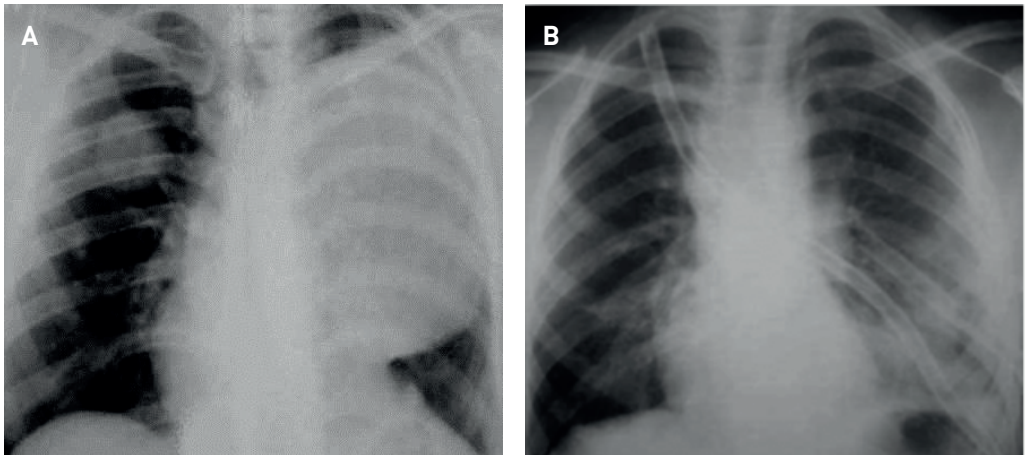


Figure 2. Bilateral pulmonary echinococcosis X-ray. A - before simultaneous two-sided echinococcectomy with transmediastinal access; B- after simultaneous two-sided echinococcectomy with transmediastinal access

Postoperative complications occurred in 17.6% of patients when applying the method by Delbe and in 18.6% when applying the method by Bobrov-Spasocucotsky. Postoperative complications occurred in 40% of the patients when applying the method by A. Vishnevsky. For patients with uncomplicated PCE, filling a cyst of fibrous capsule with vertical half purse-string Kulakeev suture resulted in postoperative complications in 22.7% of 176 (29.4%) patients. In the method of liquidation of the fibrous capsule, with capitonage carried out through a combination of horizontal and vertical sutures, no postoperative complications occurred during the 1 to 9 months after surgery.

There were no complications in the postoperative period in 9 (1.5%) patients after echinococcectomy via transmediastinal access, despite the extent of the surgical intervention: right-sided lateral thoracotomy, removal of the upper lobe of the right lung echinococcosis cyst, and subsequent removal of the echinococcosis cyst from the liver.

Discussion

Surgical intervention is the only radical method of PCEtreatment.[9,10] Important elements of surgical intervention are the technique of antiparasitic cavity treatment of the fibrous capsule and the elimination of the PCE. The means used in antiparasitic treatment of the cavity must be effective while preserving the surrounding tissues. The elimination of an echinococcosis cyst cavity of the lung should be simple, non-traumatic, and effective, regardless of the cyst size and the disease complications.[10,11]The prevention of intraoperative dissemination can be achieved by placing gauze with hypertonic saline solution (20%) or a povidone-iodine solution.[11]That's why the search for new, effective and safe methods of the echinococcal cyst cavity treatment of the lung remains relevant.[12,13] Surgery is compulsory for large cysts that are superficial, infected cysts, and cysts located in vital anatomical sites.[1,14]The surgery by A.A. Vishnevsky's method is applied in medium, large, and giant cysts when two-thirds of the cyst are above the lung surface and only one third of the cyst volume is in lung parenchyma. Whereas, small bronchiolar fistulas are treated thorough suturing by necessity, but postoperative complications often develop.[12,15]

Capitonage cystostomy is the preferred method of echinococcosis hydatid treatment. Cystostomy includes aspiration of cyst fluid and eliminating of growing membrane (Barrett's techniques).[16] Capitonage is complete closing of the cyst by suturing the cyst's wall. The method provides extra strength of lung parenchyma and prevents subsequent dissemination through air and formation of empyema.[17,18]

The primary criteria for lobectomy are cysts involving more than 50% lung lobe; festering cysts that are unresponsive to antibiotic therapy; multiple cysts that are located inside one lobe; and echinococcosis with the bronchiectasis, pulmonary fibrosis, or severe hemorrhage. The method of choice for small and medium cysts, which are located intraparenchymally, and are mostly uncomplicated and cylindrical and conical type, is a method of closed echinococcectomy, according to Delbe.[14]

In the case of multiple cysts, priority

should be given to cysts that are more likely to rupture, are of larger size, and may possibly disseminate. Large cysts require certain management of the residual space to avoid postoperative dissemination through air and formation of empyema.[19,20]

With combined PCE with involvement of abdominal organs, surgery should start from PCE, considering the possibility of high risk for developing pulmonary complications.[21] An echinococcosis cyst from the opposite lung should be eliminated with bilateral PCE, which allows a one-stage bilateral echinococcectomy of an upper lobe right liver cyst with subsequent elimination of echinococcosis cysts from the liver. The patients with combined bilateral pulmonary lesion and liver and spleen damage are good candidates for cystectomy laparotomy after bilateral phased thoracotomy and echinococcectomy of the lungs.[22] In cases of one-stage bilateral echinococcectomy thoracotomy, surgery should be started on the side of the largest cyst or with the largest threat of complications. However, this method is traumatic, and can lead to respiratory failure and increased risks of postoperative wounds in the early postoperative period. Video-assisted thoracoscopic surgery is a useful method for elimination of surface and small or moderate-sized hydatid cysts, with less morbidity compared with the usual surgery method.[23,24] To reduce surgery-related trauma, reducing the duration of the operation and the postoperative period using a bilateral one-stage sequential videoassisted thoracoscopice chinococcectomy from both lungs is effective.[23,24] The method of one-stage surgical treatment and 2-stage PCE via transmediastinal access to the eliminated right PCE and liver, through one skin incision with the use of video-assisted thoracoscopic techniques allows the reduction of trauma and the length of treatment, and patients are relieved to avoid the next-stage operations and repeated anesthesia.

This study has revealed significant proportion of complications within patients with complicated course of PCE. One of the limitations of this prospective study is the distance, because patients, with echinoccosis are pretty much about rural population, its make difficult to patients recruiting, early disease detection and monitoring of them. However, the findings highlighted the common tactic that may guide reduce of postoperative complications level and substantiate the need for further prospective studies.

Limitations. The limitation could be patients with severe comorbid pathology of the cardiovascular system, obesity, etc.

What's known? Pulmonary cystic echinococcosis, a parasitic disease, is a health care problem in developing countries. Pulmonary cystic echinococcosis has no clinical presentation and may cause impassable cough, colored sputum, hemoptysis, and fever. X-ray and Computed tomography can detect lung abscess, bronchoscopy can detect cystic lesion, and serological testing can detect antibody titer to Echinococcus granulosus.

What's new? In patients with combined PCE and abdominal organs, after thoracotomy and lung echinococcectomy, they received second-stage laparotomy and echinococcectomy from the lesser sac, from the abdomen, from the greater omentum and with a splenectomy. Also one-stage bilateral video-assisted thoracoscopic echinococcectomy was performed in patients with PCE. In the surgical treatment of echinococcosis of the lungs, preference should be given to endoscopic methods. Their use contributes to faster rehabilitation of patients without compromising the result.

Conclusion

Despite the surgical treatment success of PCE, issues of intraoperative dissemination, safety, and treatment success are still a problem. Through detailed multi-center studies the researchers will be able to best define the complications risk and relapse, choice of optimal strategies for effective surgical treatment.

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and preparation of data, primary processing of the material and their verification. N.Y., R.N.: Statistical processing and analysis of the material, writing the text of the article (material and methods, results). Sh.B., I.G, A.M., R.N.: Writing the text of the article (introduction, discussion). All authors approved the final version of the manuscript

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