

# Surgical treatment of pulmonary infection with *Echinococcus granulosus* under veno-venous extracorporeal membrane oxygenation (ECMO) – case report

*Tratamentul chirurgical al unei infecții pulmonare cu Echinococcus granulosus prin oxigenare prin membrană extracorporeală veno-venoasă*

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## Abstract

We present the case of a 51-year-old male admitted to the department of thoracic surgery for respiratory distress and suspicion of right pneumothorax. Chest and abdomen CT scan showed an abundance of cysts located in the peritoneum, liver, mediastinum and both lungs, with findings suggestive for systemic infection with *Echinococcus granulosus*, as well as extensive bilateral pulmonary emphysema and right total pneumothorax. Respiratory distress was managed by right pleural drainage without significant improvement. Right pulmonary cysts were managed by capitonnage through lateral thoracotomy. In order to resolve the left pulmonary cysts, we decided to use extracorporeal membrane oxygenation instead of one lung ventilation due to the patient's worsening condition. Veno-venous extracorporeal membrane oxygenation made possible the closure of two large cysts by capitonnage, while five smaller cysts were removed by wedge resection. The procedure was well tolerated by the patient, and on the fourth day following the procedure, extracorporeal membrane oxygenation was ceased and the patient started breathing independently.

**Keywords:** extracorporeal membrane oxygenation, *Echinococcus granulosus*

## Rezumat

În acest articol prezentăm cazul unui pacient în vârstă de 51 de ani, care a fost internat în clinica de chirurgie toracică pentru insuficiență respiratorie și suspiciune de pneumotorax drept. Examenul CT de torace și abdomen a evidențiat numeroase chisturi hepatice, pulmonare, mediastinale și peritoneale cu aspect de infecție sistemică cu *Echinococcus granulosus*. De asemenea, examenul CT a evidențiat important emfizem pulmonar bilateral și pneumotorax total drept. Pentru rezolvarea insuficienței respiratorii s-a decis practicarea pleurotomiei minime drepte, cu inserția cateterului intercostal pentru drenaj. Întrucât drenajul pleural a rezolvat pneumotoraxul drept, însă nu a determinat ameliorarea stării de sănătate a pacientului, s-a decis îndepărtarea chirurgicală a chisturilor hidatice de pe partea dreaptă prin toracotomie laterală. Pentru rezolvarea chisturilor hidatice de pe partea stângă și a posibilelor pierderi aeriene, am decis să folosim oxigenarea prin membrană extracorporeală. Prin folosirea oxigenării prin membrană extracorporeală cu abord veno-venos s-a reușit sutura a două chisturi hidatice gigante și îndepărtarea prin rezecție pulmonară atipică a altor cinci chisturi hidatice. Procedura a fost bine tolerată de pacient, iar oxigenarea prin membrană extracorporeală a fost oprită în ziua a patra postoperatorie pentru a-i permite pacientului să respire independent.

**Cuvinte-cheie:** oxigenare prin membrană extracorporeală, *Echinococcus granulosus*

## Introduction

The pulmonary infection with *Echinococcus granulosus* (EG) continues to be endemic in Romania, given its large rural population involved in sheep herding, with an incidence of 1:10,000 and a mortality of 2-4%<sup>(1)</sup>. Although in most cases the treatment is straightforward and consists in systemic chemotherapy (albendazole) and the surgical removal of the cysts membrane through various techniques depending on the involved organ, in rare cases presenting simultaneous multivisceral echinococcosis we have to consider extreme measures.

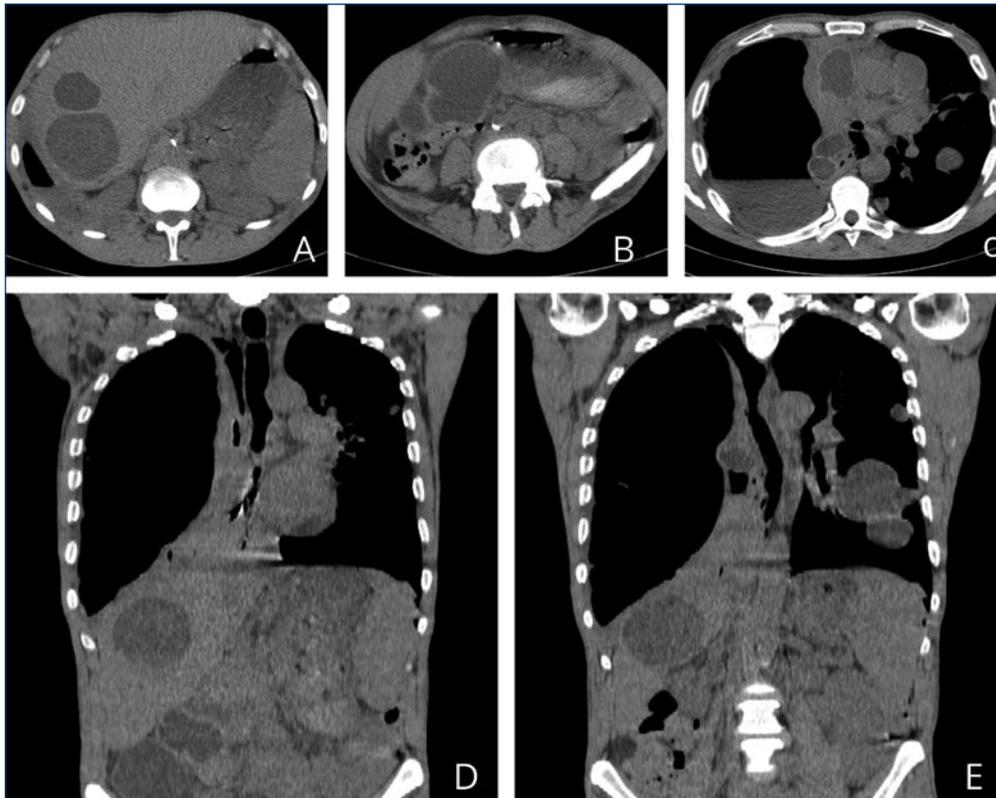
Extracorporeal membrane oxygenation (ECMO) as heart-lung bypass support is now routinely used in Intensive Care Units (ICU) for the management of pulmonary infections, severe acute respiratory distress syndrome (ARDS) or end-stage lung diseases requiring lung transplantation<sup>(2)</sup>,

but evidence supporting the utility of ECMO in pulmonary infections with *Echinococcus granulosus* is close to non-existent.

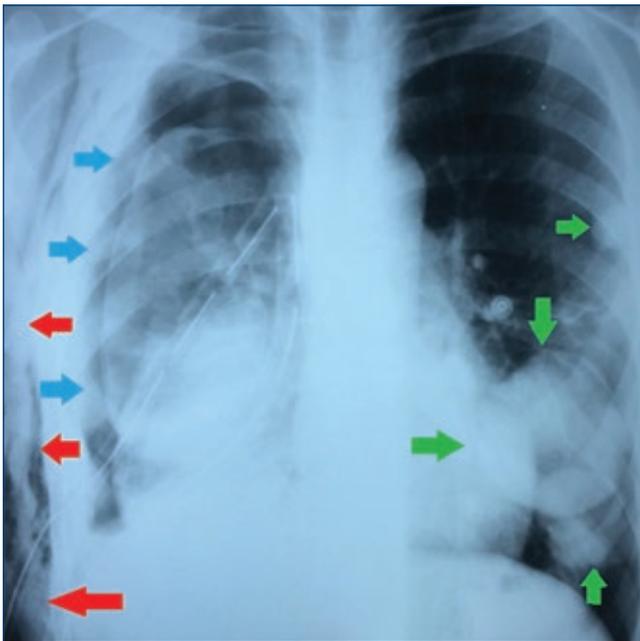
## Case report

We present the case of a 51-year-old male admitted to our clinic for respiratory distress and the suspicion of right pneumothorax. The patient had lived all his life in Argeș county and worked as a shepherd at the time of presentation, he was a smoker (60 packs-year), he consumed large quantities of alcohol and up to this moment his medical history was unremarkable.

The patient was examined in our emergency room, at which time he presented dyspnea, dry cough, enlarged liver and peripheral oxygen saturation of 88%, which corrected to 92% under oxygen 3 liters/minute. The ini-



**Figure 1.** CT scan of the abdomen and chest: A) hepatic cysts; B) peritoneal cysts; C) right pneumothorax, right pulmonary cysts, mediastinal cyst; D) large hepatic cyst, and E) left pulmonary cysts



**Figure 2.** Chest X-ray performed after surgery: on the right lung: partially expanded right lung surrounded by air (blue arrows), cutaneous emphysema of the thorax (red arrows) and three cysts visible in the left lung (green arrows)

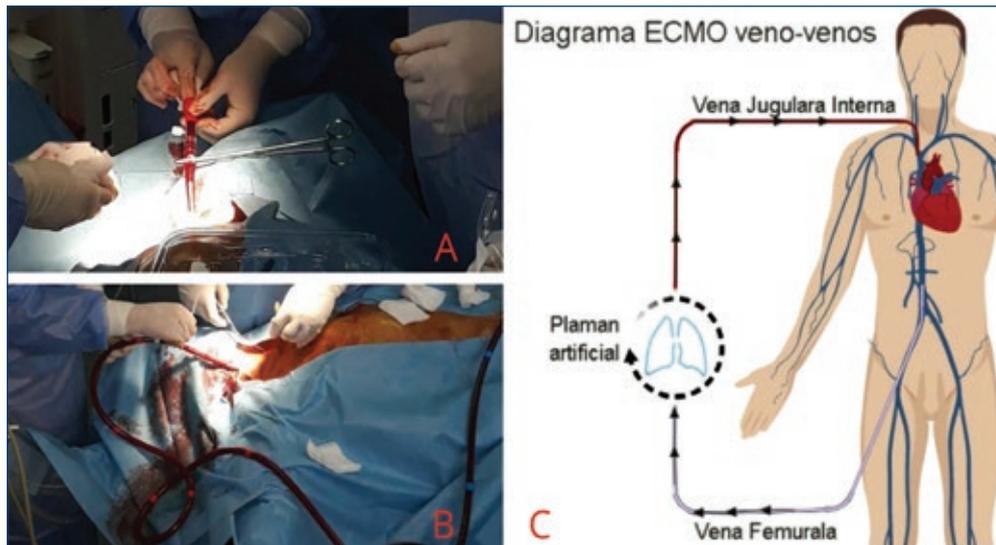
tial chest X-ray showed right-sided pneumothorax and multiple opacities of different sizes, located on the left pulmonary field. Following a difficult anamnesis, we discovered that the patient had vomited a clear liquid a day before and coughed up white fragments of soft tissue.

Detailed full body CT scan showed an abundance of cysts located in the peritoneum, liver, mediastinum and both lungs, some intact, while others without content, findings which were suggestive for a systemic infection with *Echinococcus granulosus* in the context of the patient's symptomatology and anamnesis. Extensive left pulmonary emphysema and right total pneumothorax were also visible during the scan (Figure 1).

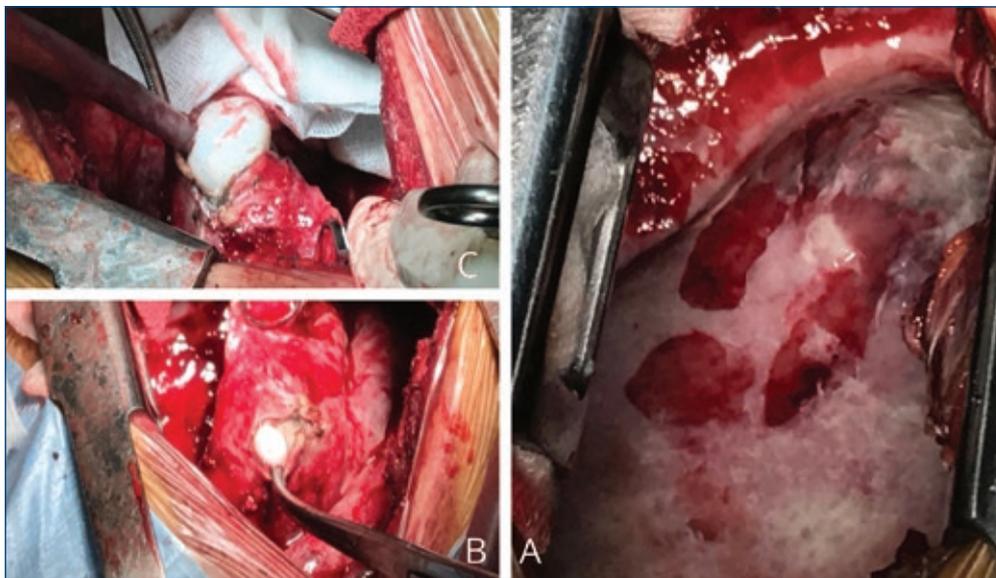
The patient's condition quickly deteriorated, with hypoxia present, which forced us to perform right pleural drainage with a 28 Fr catheter in order to resolve the respiratory distress. Despite chest drainage, the patient's condition remained unstable due to the poor state of the pulmonary parenchyma (important bilateral emphysema) and massive air leakage.

Following the discussion of the case by a medical council comprising of pulmonologists, thoracic surgeons, general surgeons, anesthesiologists and infectious physicians, a conclusion was reached that the treatment alone with albendazole and antibiotics will prove ineffective and restoring compromised pulmonary function by removing the cyst and suturing the air leaks were critical.

The first step was to remove the right pulmonary cysts and resolve the persistent air leaks in an emphysematous lung by muscle sparing right lateral thoracotomy. The procedure was successful in resolving the cysts, but the patient could not withstand one lung ventilation (OLV) during the operation and forced us to alternate periods of apnea with periods of ventilation, which made suturing the air leaks a challenge. Pulmonary lobectomy was not an option given the patient's critical state. Following surgery, the patient continued to present air leakage and still required mechanical ventilation (Figure 2).



**Figure 3.** Operative images of the cannulation procedure and ECMO diagram by Mr. Mihai Dumitrescu: A) the shorter arterial cannula is inserted in the internal jugular vein before ECMO; B) ECMO has been started and oxygenated blood is flowing through the arterial cannula placed in the internal jugular vein; C) diagram of veno-venous ECMO showing RECOVERY from femoral vein (white color) and FLOW into internal jugular vein (red color)



**Figure 4.** Operative images of the left lung: A) partially collapsed left lung which is covered by a thick visceral pleura; B) pneumotomy reveals the membrane of a cyst; C) removal of the cyst membrane

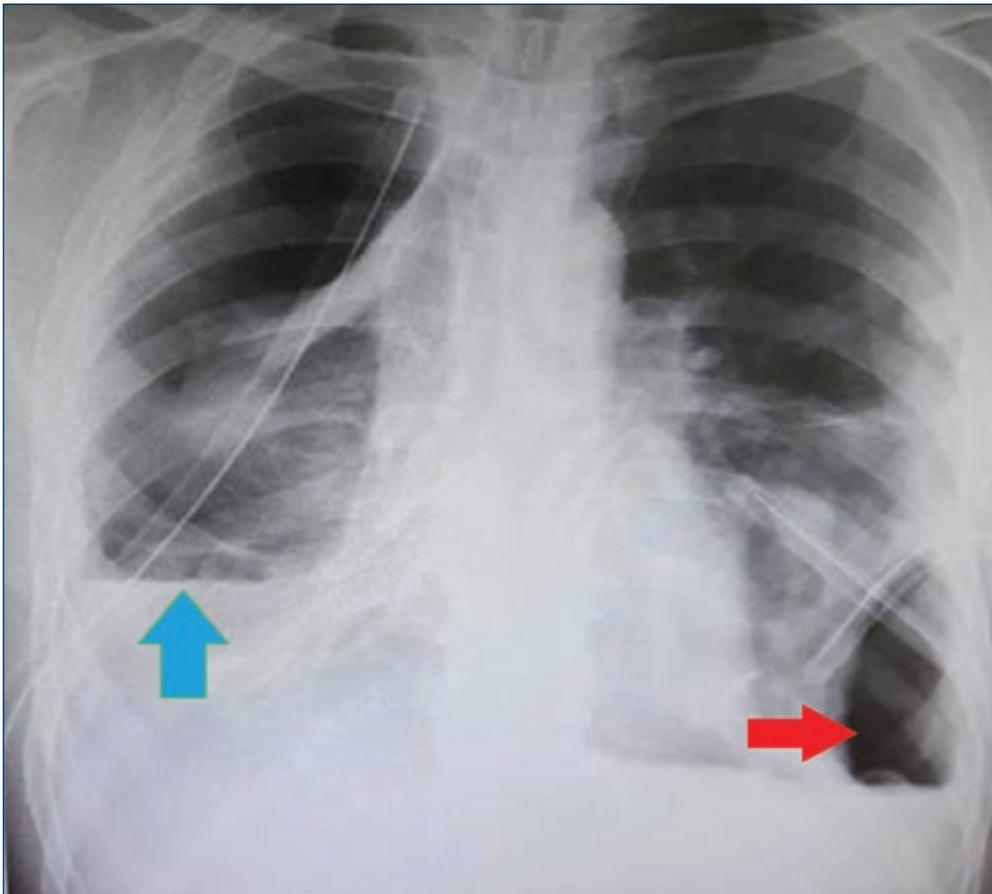
In order to resolve the cysts on the left side in a patient who could not withstand another procedure under OLV due to the poor state of the pulmonary parenchyma, we decided to attempt using ECMO as bypass for the pulmonary function.

With the patient positioned in dorsal recumbent position, a central venous line and an arterial line in place, we opted for veno-venous ECMO by inserting a 21 Fr arterial cannula in the right internal jugular vein (IJV) for the flow and a 23 Fr venous cannula in the right femoral vein (FV) for recovery. The cannulas were inserted without ultrasound guidance for a maximum length of 30 cm in the IJV, in order to reach the right atrium, and 60 cm in the FV to position below the hepatic veins. Given the patient's weight of 65 kg and his height of 175 cm, we decided on a flow of 3000 mL/min, which was monitored for 10 minutes before ceasing mechanical ventilation (Figure 3).

Ten minutes later, the patient was repositioned in lateral recumbent position, the mechanical ventilation was ceased and left muscle sparing lateral thoracotomy,

in the fifth intercostal, was performed. Upon entry in the pleural cavity, the lung was manually deflated before careful inspection. After removing a part of the thick visceral pleura, two large cysts of the inferior left lobe were discovered and closed by capitonnage after removal of the membranes (Figure 4). Five cysts of the superior left lobe were also discovered and removed by wedge resection, given their small size and disposition. The procedure lasting 155 minutes was well tolerated by the patient and the ECMO flow was unaffected by the lateral decubitus position.

After surgery, the patient remained sedated, intubated, but not ventilated, and under ECMO support for three days. From the third day, the patient was conscious, able to breath on his own, with minor air leakage present bilaterally, and ECMO support was turned down to a minimal 200 mL/min flow every four hours for periods lasting 10-15 minutes in order to ease the cessation from ECMO. On the fourth day following the procedure, ECMO was ceased definitely and the patient was able to breath on his own (Figure 5).



**Figure 5.** Chest X-ray performed after removal of ECMO: the inferior left lobe has incomplete basal expansion (red arrow) and pleural fluid is present on the right side (blue arrow)

## Discussion

Although lung transplantation remains the main indication for ECMO in thoracic surgery, recently its use has also been reported for the surgery of tracheal tumors, tracheal or bronchial fistulas and pulmonary resections in trauma patients<sup>(3)</sup>.

In pneumology, the main indication for ECMO remains the treatment of ARDS, especially in infections with influenza or in pneumococcal pneumonia, but we haven't found any reports recommending ECMO for treating the complications of echinococcosis<sup>(4)</sup>.

We did find a case in which ECMO was used to treat ARDS in a patient with a ruptured pulmonary cyst and sepsis<sup>(5)</sup>, but our case is the first operated successfully under ECMO during which multiple wedge resections were performed and cavities were repaired by capitonnage.

Our rationale for using ECMO was to be able to perform the procedure in a patient under respiratory distress, notwithstanding OLV, while at the same time refrain from using mechanical ventilation, which was partially responsible for the persistent air leaks, and to allow for natural healing of the smaller pulmonary fistulas. Currently, after the patient's pulmonary function has stabilized, ECMO is ceased gradually, while mechanical ventilation is reintroduced. This was not an option for us since mechanical ventilation promoted air leakage, so we were forced to cease ECMO at a later stage.

Still, several questions were raised before and after the procedure, such as: does ECMO increase the risk of liver cysts rupture and parasite dissemination, does the use of ECMO contribute to faster closing of the air leaks, and is

ECMO indicated in multivisceral advanced disease, with poor outcome, given its high costs?

Although fellow practitioners could argue that this case is an "end-stage" in the sense that the disease is borderline untreatable and the patient presented important bilateral emphysema, we cannot disconsider the patient's right to medical treatment and it is the authors' opinion that ECMO was the patient's only option for surgical treatment.

## Acknowledgements

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