Pneumonektomija zaradi velike simptomatske hidatidne ciste

Giant symptomatic hydatid cyst requiring pneumonectomy

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Izvleček

Namen: *Prispevek prikazuje diagnostično obravnavo, zdravljenje in nekatere zaplete zdravljenja bolnikov s hidatidno boleznijo pljuč. Cilj operativnega zdravljenja je odstranitev ciste in ohranitev zdravega pljučnega parenhima. V primeru preraščanja pljučne arterije, ki je sicer redko, pa je včasih potrebna odstranitev pljučnega krila, kar je povezano z možnostjo težkih pooperativnih zapletov.*

Poročilo o primeru: *Prikazan je primer 27 letne bolnice, pri kateri je bila zaradi preraščanja pljučne arterije s hidatidno cisto potrebna odstranitev levega pljučnega krila. Po operaciji je prišlo do razvoja akutnega respiratornega distres sindroma (ARDS) preostalega desnega pljučnega krila. Potrebna je bila zunajtelesna membranska oksigenacija (ECMO). Kljub resnemu zapletu obsežne kirurške resekcije je bol-*

Abstract

Purpose: *This paper discusses the diagnostic approach, treatment and certain complications in the treatment of patients with hydatid lung disease. The objective of surgical treatment is removal of the cyst and preservation of the healthy lung parenchyma. In cases of overgrowth of the pulmonary artery, which are rare, it is sometimes necessary to remove the lung;this is linked with the possibility of serious postoperative complications.*

Case report: *We present the case of a 27–year–old patient with a hydatid cyst overgrowing the pulmonary artery and requiring removal of the left lung. After the surgery, acute respiratory distress syndrome (ARDS) developed in the remaining right lung. Extracorporeal membrane oxidation (ECMO) was required. Despite a serious complication of the extensive surgical resec-*

nica po štirih tednih brez večjih težav zapustila bolnišnico.

Zaključek: *Pri velikih hidatidnih cistah pljuč je kljub velikosti v večini primerov možna operacija z ohranitvijo pljučnega tkiva in dodatna terapija z Albendazolom. Odstranitev pljučnega krila zaradi preraščanja pljučne arterije pa lahko bistveno poslabša pooperativni potek zdravljenja. Pri planiranju operativnega posega je bistven natančen slikovni prikaz struktur pljučnega hilusa.*

tion, the patient left the hospital after four weeks without major health problems.

Conclusion: *In patients with giant pulmonary hydatid cysts it is possible to perform surgery, preserve the lung and continue treatment with the addition of albendazole therapy. The removal of the lung, on account of overgrowth of the pulmonary artery, can significantly affect the course of post-operative treatment. When planning the surgical procedure, a precise image of the pulmonary hilar structures is essential.*

INTRODUCTION

Hydatid disease is a parasitic disease, caused by the dog tapeworm Echinococcusgranulosus. It is present throughout the world and is truly endemic in the Mediterranean countries, South America, Australia and Eastern Africa (1). In adult humans,hydatid cysts develop most commonly in the liver (more than 65%) and lungs (25%), whereas other organs (heart, muscles, brain, kidneys, spleen) are affected rarely (1, 2). Pulmonary hydatidcysts may become infected or calcified; they can also perforate and cause an anaphylactic reaction and larger pleural effusion (1). Pulmonary hydatid cysts larger than 10 mm are categorized as giant cysts (3).

An adult Echinococcus tapeworm is 7mm long, is hermaphrodite, and its body is divided into several reproduction units (proglottids), the last one being gravid (1). Echinococcus is transmitted orally and via faeces and requires an intermediate and definitive host. A dog which eats an organ from an intermediate host (sheep, rodent, pig) becomes the definitive host of an adult tapeworm. Adult E. granulosus release eggs within the canine intestine which will be transported out of the body via the faeces. When contaminated waste is excreted into the environment, an intermediate host has the potential to contract the parasite, perpetuating the cycle. Humans can also be an intermediate host for E. granulosus.

The larval tapeworm (oncosphere) penetrates the intestinal wall and enters the portal bloodstream or lymph and then the liver, lungs, nervous system and other organs (1). In a few days, anechinococcalcyst (metacestode) develops from the oncosphere (in humans most commonly in the liver or lungs), and by growing creates pressure on the neighbouring tissues. The fundamental principles of treatment are the use of albendazole (4), and in cases of giant cysts or complications a classic or minimally invasive surgical procedure (5).

CASEREPORT

A 27–year–old female patient was admitted to the Department of Pulmonary Diseases of the University Medical Centre Maribor on account of a chesty cough she had been experiencing for some months. Chest–CT (computed tomography) revealed a cystic formation 18.7x11x15 cm in size in the upper left pulmonary lobe (Figure 1). Echinococcus infection was confirmed by serology. Owing to deterioration in the patient's state of health, including increase in body temperature, early signs of sepsis, and radiologically confirmed involvement of the left lung, she was transferred to the Department of Thoracic Surgery, where she received surgical treatment. The patient had already received albendazole.

Figure 1. *Chest–CT with giant hydatid cyst of the upper left pulmonary lobe.*

Figure 3. *Aspirating the contentsof the hydatid cyst.*

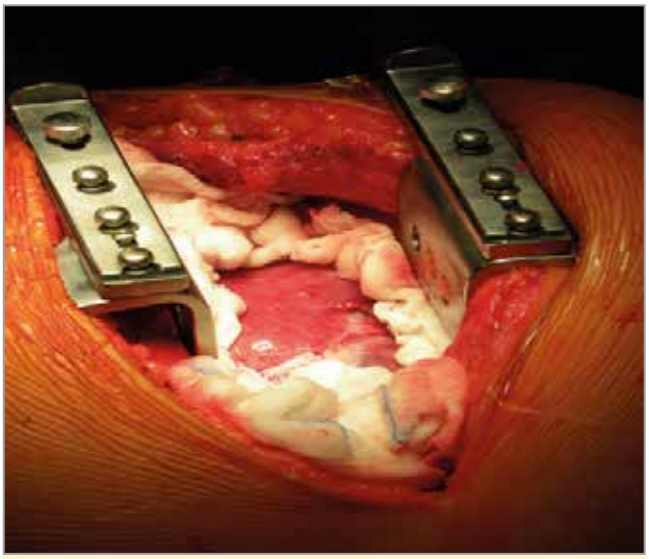

Figure 2. *Left pulmonary hydatid cyst covered with compresses soaked with hypertonic saline solution.*

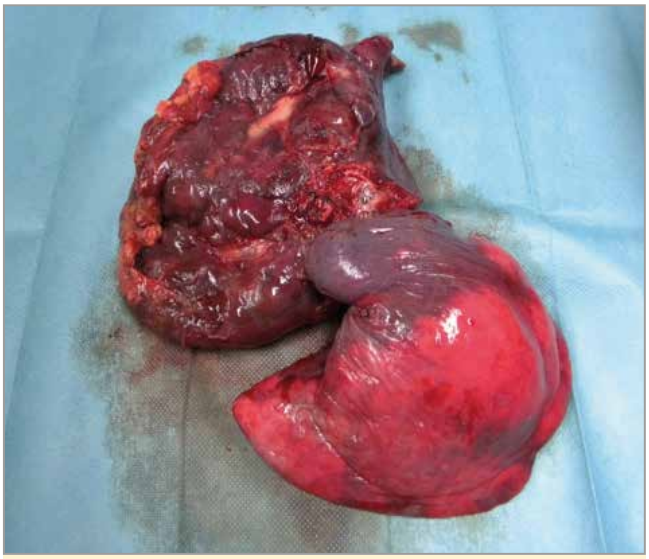

Figure 4. *Removed left lung with aspirated giant hydatid cyst.*

After short and intensive preoperative preparation, coordinated by an infectious disease specialist and anaesthesiologist, the pleural space was explored through a left posterolateral thoracotomy. A massive hydatid cyst with numerous inflammatory adhesions to the parietal pleura was observed in the left upper lung lobe. The left lower lobe and pulmonary hilus were overgrown by the cyst. It was impossible to remove the cyst whilepreserving the pulmonary parenchyma; therefore,it was decided to perform left upper lobectomy. The pleural space was covered with compresses soaked in hypertonic saline to protect the surgical field in case of cyst rupture and to prevent dissemination or anaphylactic reaction (Figure 2). The cyst contentswere aspirated (Figure 3) and dissection of the left hilar structures was begun. The main pulmonary artery was found to be overgrown by the cyst, and therefore left pneumonectomy was performed. The stump of the left main bronchus was additionally buttressed.The left lung and the cyst were sent for histopathology (Figure 4).

Postoperatively, the patient was transferred to the perioperative intensive care unit. the patient's health

deteriorated on day 1 post–surgery. She developed a right–sided pneumonia and acute respiratory distress syndrome (ARDS). The patient required extracorporeal membrane oxidation (ECMO), and therefore she was transferred to the Department of Internal Intensive Medicine of the University Medical Centre Ljubljana. After 1 week she was re–admitted to our department. She recovered well and was discharged 4 weeks after surgery.

DISCUSSION

Hydatid disease does not represent a major health issue in Slovenia, where the yearly incidence is 0.34 per 100,000 people (2). Many diagnoses are made in immigrant patients, as was the case in our patient.
The liver is the most commonly affected organ, followed by the lung. Hydatid disease occurs more frequently in the right lung and in the lower half of the lung (6). Patients can be asymptomatic for a considerable time. When the growing cyst starts to put pressure on the lung parenchyma, discomfort,a chesty cough, chest pain and heavy breathing occur. Rupture of a cystis a serious adverse event, causing anaphylactic reaction, extensive pleural effusion, and cyst infection with septic symptoms (7), and these are the main clinical symptoms indicating a deterioration in the patient's condition.

To establish the diagnosis, diagnostic imaging and serologic testing are required. Hydatid cyst needs to be distinguished from benign cysts, cavitarytuberculosis (TB), mycoses, abscesses and benign or malignant neoplasms. Following conventional X–ray imaging, a chest–CT precisely defines the size and contents of the cyst, as well as its relation to surrounding structures. Moreover, MRI (magnetic resonance imaging) should be performed to reveal possible overgrowth of hilar structures (8). In our case, MRI was not performed, because we could find no guidelines or reports of its beneficial use in such cases. However, MRI is a useful preoperative diagnostic tool whenever there is a substantial rearrangement of the lung hilum due to a tumor. Antibodies against echinococcal antigens were demonstrated by serologic testing (9). Both examinations may be improved by demonstrating protoscolex or echinococcal DNA in a sample obtained by fine needle aspiration, or in sputum (9). Scolicidal therapy can be introduced as single or supportive therapy; the treatment of choice is albendazoleat 10–15 mg/kg a day for 3–6 months (1).

Surgical treatment plays a crucial role when treating giant hydatid cysts, and covers a wide range of possible complications. There are many options for surgical resection; however, the main goal is safe removal of the intact cyst and preservation of the healthy surrounding pulmonary parenchyma (10).

The major problem of surgical treatment and postoperative complications remains the inflammatory or direct overgrowth of hilar structures, and the pulmonary artery in particular. In such cases, good preoperative diagnostic imaging is crucial. This patient had a rare complication of pneumonectomy; ARDS was promoted by the severe inflammatory involvement of the lung parenchyma and poor preoperative status. An arterial perfusion defect or its deformation points to arterial involvement and dictates the necessary preparation for surgical resection, including the use of ECC (extra–corporeal circulation) (11). In order to achieve completeexcision of the cystin our patient, removal of the left lung was required. This is often the case when large tumours are encountered which prevent clear access to the lung hilum. The patient's condition prevented a more extensive approach and longer surgical procedure, but even so she suffered major pre– and postoperative complications.

CONCLUSION

Classic or minimally invasive surgical procedures are the method of choice in treating giant hydatid pulmonary cysts or their complications. Preoperative diagnostic procedures, especially diagnostic radiology, significantly affect the planning of the necessary surgical procedure.

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