

LAPAROSKOPSKO LEČENJE PROSTE DŽINOVSKE CISTE U JETRI

PRIKAZ SLUČAJA

CASE REPORT

LAPAROSCOPIC MANAGEMENT OF A GIANT SIMPLE HEPATIC CYST

Vuk Aleksić¹, Vladimir Špica², Zorana Bokun-Vukašinović³,
Radmila Ćulafić⁴, Svetlana Mijatović¹, Nebojša Mitrović²

¹ Služba neurohirurgije, Kliničko bolnički centar „Zemun“, Beograd, Srbija

¹ Neurosurgery Department, “Zemun” Medical Center, Belgrade, Serbia

² Služba hirurgije, Kliničko bolnički centar „Zemun“, Beograd, Srbija

² Surgery Department, “Zemun” Medical Center, Belgrade, Serbia

³ Služba patologije, Kliničko bolnički centar „Zemun“, Beograd, Srbija

³ Pathology Department, “Zemun” Medical Center, Belgrade, Serbia

⁴ Stomatološki fakultet, Univerzitet u Beogradu, Beograd, Srbija

⁴ Faculty of Dental Medicine, Belgrade, Serbia

SAŽETAK

Uvod: Urođene proste ciste u jetri su izuzetno retke i obično su asimptomatske. Najbolji terapijski pristup za lečenje prostih cista u jetri još uvek je predmet naučnih rasprava.

Prikaz slučaja: Prikazujemo slučaj gigantske ciste u jetri uklonjene minimalno invazivnim laparoskopskim pristupom kod žene stare 68 godina, kod koje se bolest prezentovala brzim gubitkom u telesnoj masi.

Zaključak: Usled nedostatka randomizovanih kliničkih studija koje pored rezultate minimalno invazivnog (laparoskopskog) i otvorenog hirurškog pristupa u lečenju cista u jetri, savetujemo personalizovani medicinski pristup nakon sagledavanja karakteristika ciste, ali i pacijenta.

Ključne reči: cista u jetri, urođena cista, laparoskopija

ABSTRACT

Introduction: Congenital simple hepatic cysts are extremely rare and are usually asymptomatic. The best therapeutic approach to treating simple liver cysts remains the subject of scientific debate.

Case report: We present the case of a giant liver cyst removed with a minimally invasive, laparoscopic approach in a 68-year-old woman, who presented with rapid weight loss.

Conclusion: Due to the lack of randomized clinical trials comparing minimally invasive (laparoscopic) and open surgery approaches in liver cyst treatment, we recommend a personalized medical approach, applied after analyzing the characteristics of both the cyst and the patient.

Keywords: hepatic cyst, congenital cyst, laparoscopy

Autor za korespondenciju:

Vuk Aleksić

Služba neurohirurgije, Kliničko bolnički centar „Zemun“

Vukova 9, 11000 Beograd, Srbija

Elektronska adresa: aleksicvuk@hotmail.com

Corresponding author:

Vuk Aleksić

Neurosurgery Department, “Zemun” Medical Center

9 Vukova Street, 11000 Belgrade, Serbia

E-mail: aleksicvuk@hotmail.com

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UVOD

Ciste u jetri su, po pravilu, solitarne neparazitske lezije jetre, poznate i kao proste ciste u jetri. Poreklo prostih cista u jetri još uvek nije poznato, ali se čini da su urođene. Ove ciste su izuzetno retke [1,2]. Takođe su obično bez kliničkih simptoma. Izuzetno velike, džinovske ciste mogu postati simptomatske usled rupture, efekta mase, krvarenja, i/ili infekcije. One takođe mogu izazvati atrofiju tkiva jetre, ponekad uz kompežatornu hipertrofiju nezahvaćenog dela jetre [3]. Terapijski pristup lečenju prostih cista u jetri je još uvek predmet naučne debate [4]. Terapija može uključivati: perkutanu aspiraciju iglom, ubrizgavanje različitih sklerozirajućih agenasa, fenestraciju laparoskopskim putem ili putem otvorene hirurgije, hiruršku cistektomiju ili marsupijalizaciju [1].

Ovde predstavljamo slučaj džinovske ciste u jetri uklonjene minimalno invazivnim laparoskopskim pristupom, kod šezdeset osmogodišnje pacijentkinje kod koje se bolest manifestovala naglim gubitkom telesne mase.

PRIKAZ SLUČAJA

Šezdeset osmogodišnja pacijentkinja javila se u našu hitnu službu žaleći se na osećaj nelagodnosti u trbuhu. Pacijentkinja je prijavila značajan gubitak telesne mase od 17 kg u tri meseca. Na pregledu je abdomen bio blago nadut a prednji trbušni zid je bio iznad nivoa grudnog koša. Indirektna perkusija prstom o prstom u projekciji jetre proizvela je tup zvuk. Pacijentkinja je demonstrirala minimalnu osetljivost na palpaciju, ali nije bilo mišićnog defansa. Takođe, nije bilo znakova žutice, ascita ili edema nogu. Osim minimalno uvećanog nivoa aspartat aminotransferaze (AST), laboratorijski nalazi su bili u fiziološkim granicama. Nivoi tumor markera bili su takođe u okviru normalnih referentnih vrednosti. Nalaz antitela na ehinokokus je takođe bio negativan. Obavljen je ultrazvuk abdomena koji je pokazao džinovsku

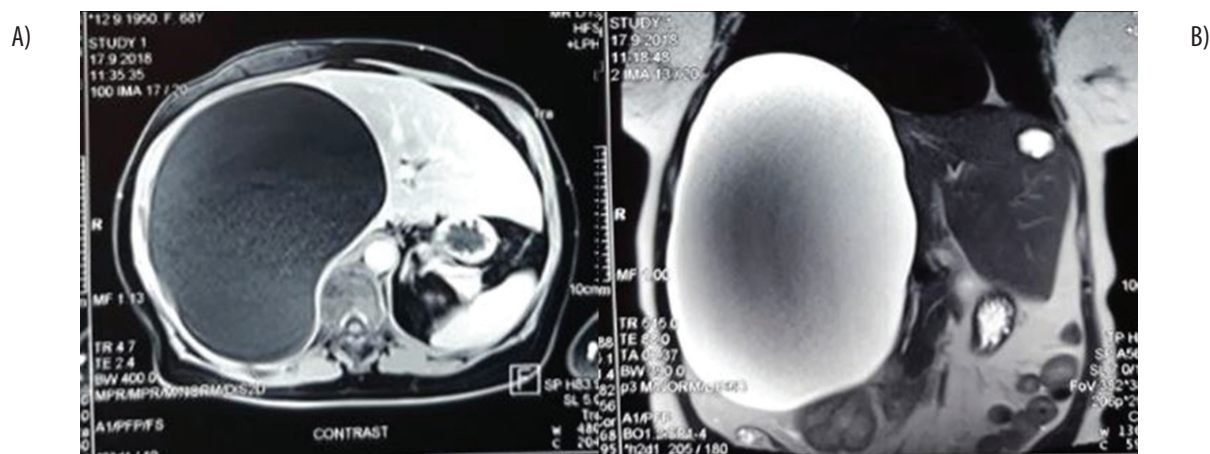
INTRODUCTION

A hepatic cyst is typically a solitary non-parasitic lesion of the liver, also known as a simple hepatic cyst. The origin of simple hepatic cysts is still unknown, but it appears that they are congenital. These cysts are exceedingly uncommon [1,2]. Also, they are usually without clinical symptoms. Extremely large, giant cysts can become symptomatic due to rupture, mass effect, hemorrhage, and/or infection. They can also produce liver tissue atrophy, sometimes with compensatory hypertrophy of the unaffected liver part [3]. The therapeutic approach for simple liver cysts is still the subject of scientific debate [4]. Therapy may include percutaneous needle aspiration, injection of different sclerosing agents, laparoscopic or open surgery fenestration, surgical cystectomy or marsupialization [1].

We present the case of a giant liver cyst removed with a minimally invasive, laparoscopic approach in a 68-year-old woman, who presented with rapid weight loss.

CASE REPORT

A 68-year-old female patient presented to our emergency department with abdominal discomfort. The patient reported significant weight loss of 17 kg in three months. On physical examination, the abdomen was slightly distended, and the anterior abdominal wall was above the level of the chest. Indirect finger percussion of the liver span produced a dull sound. There was minimal abdominal palpation tenderness, but no muscular defense. Also, there were no signs of jaundice, ascites, or edema of the legs. With the exception of a minimally elevated aspartate aminotransferase (AST) level, laboratory findings were within the physiological range. Tumor marker levels were also within normal ranges. The echinococcus antibody detection finding was negative. Abdominal ultrasound was performed and showed a giant, anechoic, oval-shaped, sharply edged cyst, which occupied the



Slika 1. NMR snimak abdomena na kojem se vidi velika cista u jetri. (A) T1 NMR sekvenca, transverzalna ravan. (B) T2 NMR sekvenca, koronalna ravan.

Figure 1. Abdominal MRI showing the large liver cyst. (A) T1-weighted imaging MRI sequence, transverse plane. (B) T2-weighted imaging MRI sequence, coronal plane.



Video1. https://casopis.rklbg.org.rs/images/2020/Video_1.mp4

Video2. https://casopis.rklbg.org.rs/images/2020/Video_2.mp4

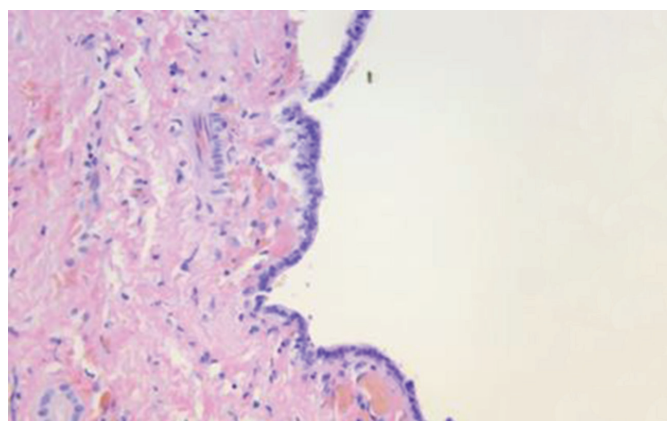


Slika 2. Cista nakon ekstrakcije pored šprica od 25 ml. (Cista je prethodno aspirirana)

Figure 2. Cyst after extraction next to a 25 ml syringe. (The cyst was previously aspirated.)

anehoičnu ovalnu cistu oštih ivica, koja je zauzimala čitav desni režanj jetre. Dimenzije ciste nisu mogle da budu utvrđene ultrazvukom. Snimanje abdomena nuklearnom magnetnom rezonancom (NMR) pokazalo je cističnu masu dimenzija 200 mm x 260 mm x 260 mm u desnom režnju jetre, koja se prostirala distalno u abdomen i potiskivala je donju šuplju venu, bez znakova tromboze ili infiltracije. T1 sekvenca magnetne rezonance pokazala je veoma nizak intenzitet homogenog signala ciste, dok je T2 sekvenca pokazala povećan intenzitet signala, bez pojačanja intenziteta signala posle primene kontrasta. Cista je takođe potiskivala i duodenum i glavu pankreasa, kao i desni bubreg, kaudalno (Slika 1).

Pacijentkinja je elektivno operisana. Primenjen je laparoskopski pristup pomoću tri trokara. Nakon kreiranja pneumoperitoneuma, putem pupčanog otvora, obavljena je vizuelizacija ciste. Sledeći korak bila je perkutana aspiracija ciste putem koje je sakupljeno oko 6 litara serozne tečnosti. Cistektomija je potom obavljena harmonijskim hirurškim makazama (Video1); tokom cistektomije obavljena je parcijalna incizija zida ciste i izvršena je ekstrakcija kroz pupčani otvor (Video 2) (Slika 2). Međutim, intrahepatični deo zida ciste, koji je bio blisko srastao sa hepatobilijarnim vaskularnim elementima, ostao je intaktan.



entire right liver lobe. The dimensions of the cyst could not be determined. Abdominal magnetic resonance imaging (MRI) was performed and revealed a 200 mm x 260 mm x 260 mm cystic mass in the right liver lobe, which extended distally into the abdomen and compressed the inferior vena cava, without signs of thrombosis or infiltration. The T1-weighted imaging MRI sequence showed that the cyst had very low homogeneous signal intensity, while the T2-weighted imaging MRI sequence showed increased signal intensity, without enhancement of the signal intensity after contrast administration. The cyst also compressed the duodenum and the pancreas head, as well as the right kidney, caudally (Figure 1).

The patient was operated on, electively. The laparoscopic approach was used with three trocars. After creating pneumoperitoneum, via the umbilical port, visualization of the cyst was performed. The next step performed was percutaneous aspiration of the cyst, and approximately 6 liters of serous fluid was collected. Cystectomy was then performed with harmonic surgical scissors (Video1); during the cystectomy, a partial excision of the cyst wall was performed and it was extracted through the umbilical port (Video 2) (Figure 2). However, the intrahepatic part of the cyst wall adherent to the hepatobiliary vascular elements was left intact.

Slika 3. Patohistološki nalaz na kojem se vidi prosta urođena cista formirana od bilijarnih duktusa (hematoksilin-eozin, uveličanje x400).

Figure 3. Pathohistological finding showing the simple congenital cyst formed from the bile duct (hematoxylin-eosin, magnification x400).

Svi otvori su zatvoreni manuelno. Postoperativni period je prošao bez komplikacija, i pacijentkinja je otpuštena tri dana od operacije. Patohistološki nalaz pokazao je prostu urođenu cistu nastalu iz bilijarnog duktusa (Slika 3). Na prvoj kontroli, mesec dana nakon operacije, pacijentkinja se osećala dobro, nije se žalila ni na šta, i povratila je oko tri kilograma telesne mase. Nakon ovog kontrolnog pregleda pacijentkinja više nije praćena.

DISKUSIJA

Na osnovu ultrazvučnih pregleda, prevalenca cista u jetri je oko 2,5%. Međutim, tačna učestalost pojave cista u jetri nije poznata pošto je većina njih asimptomatična. Ciste u jetri su klasifikovane u četiri glavne grupe, na osnovu porekla njihovog nastanka, na: urođene, neoplastične, upalne i traumatske. Proste urođene ciste su uglavnom solitarne i veruje se da nastaju kao posledica abnormalnog razvoja intrahepatičnih bilijarnih duktusa [5,6]. U slučaju koji je ovde predstavljen, cista je bila solitarna, unilokularna, i, na osnovu patohistološkog nalaza, prema poreklu, urođena.

Proste ciste variraju u prečniku od nekoliko milimetara do preko 20 centimetara. Najveća cista opisana u literaturi sadržala je 17 litara serozne tečnosti [7]. U slučaju koji je ovde predstavljen, cista je bila masivna i sadržala je oko 6 litara serozne tečnosti.

Većina prostih urođenih cista su asimptomatske, ali mogu postati simptomatske i može doći do komplikacije usled rupture, intracističnog krvarenja, ili sekundarne bakterijske infekcije. Ponekad potiskivanje susednih struktura može izazvati specifične simptome, kao što su edem donjih ekstremiteta (usled pritiska na donju šuplju venu), portalna hipertenzija i njene komplikacije (usled pritiska na portalne vene), iliolestaza (usled pritiska na bilijarno stablo) [5,8,9]. Ponekad, masivne ciste mogu izazvati bolove u trbuhu, pre svega u desnom gornjem kvadrantu abdomena, usled istežanja kapsule jetre. Retko se može uočiti ili napipati masa u abdomenu. U slučaju koji je ovde predstavljen, pacijentkinja je osećala nelagodnost u trbuhu i prijavila je značajan gubitak telesne mase u periodu od tri meseca pre prijema u bolnicu. Abdomen je bio blago nadut, a pacijentkinja je pokazala minimalnu osteljivost na palpaciju abdomena.

Radiološko snimanje je korisno za otkrivanje i utvrđivanje karakteristika cista u jetri. Snimci cista u jetri kompjuterizovanom tomografijom (CT) pokazuju atenuaciju vode, po pravilu hipoatenuaciju od oko 0-10 Hounsfieldovih jedinica. Tipičan NMR nalaz ove vrste ciste pokazuje isti intenzitet signala kao i voda, zid ciste je obično nevidljiv, i signal se ne pojačava posle ubrizgavanja intravenskog kontrasta. Isto važi i za

All port sites were closed manually. The postoperative period was uneventful, and the patient was discharged on the third postoperative day. The pathohistological finding showed a simple congenital cyst originating from the bile duct (Figure 3). At the first check-up, one month after surgery, the patient was feeling well, she had no complaints, and she regained about three kilograms. After this point the patient was lost to follow-up.

DISCUSSION

The prevalence of liver cysts is about 2.5%, based on ultrasound examinations. However, the precise frequency of hepatic cysts is not known since most of them are asymptomatic. Hepatic cysts are classified into four main groups, based on the origin of formation: congenital, neoplastic, inflammatory, and traumatic. Simple congenital cysts are mainly solitary, and are believed to arise from the abnormal development of intrahepatic bile ducts [5,6]. In the presented case, the cyst was solitary, unilocular, and, according to pathohistological findings, it was congenital in origin.

Simple cysts vary in diameter from a few millimeters to more than 20 centimeters. The largest cyst reported in literature contained 17 liters of serous fluid [7]. In the presented case, the cyst was massive, and contained approximately 6 liters of serous fluid.

The majority of simple congenital cysts are asymptomatic, but they can become symptomatic and complicated due to rupture, intracystic hemorrhage or secondary bacterial infection. Sometimes, compression of adjacent structures can cause characteristic symptoms, such as edema of the lower extremities (due to inferior vena cava compression) portal hypertension and its complications (due to the compression of the portal veins), or cholestasis (due to biliary tree compression) [5,8,9]. Sometimes, massive cysts can cause abdominal pain, mainly in the right upper abdominal quadrant, due to the stretching of the liver capsule. Rarely, a palpable mass or an abdominal lump can be detected. In the case presented here, the patient experienced abdominal discomfort and reported significant weight loss in the three months prior to admission. The abdomen was slightly distended and there was minimal abdominal palpation tenderness.

Radiological imaging is useful for detection and determination of the characteristics of hepatic cysts. Computed tomography (CT) scans of liver cysts show water attenuation, typically homogeneous hypo-attenuation of about 0-10 Hounsfield units. A typical MRI finding of this type of cyst demonstrates the same signal intensity as water; the cyst wall is usually imperceptible; its signal does not increase in intensity after intravenous contrast material administration. The same applies to a

snimanje takve ciste na skeneru. Stoga, T1 sekvenca pokazuje uniformni signal veoma niskog intenziteta, dok T2 sekvenca pokazuje pojačanje intenziteta signala veće nego druge T2 hiperintenzne lezije jetre, kao što je hemangioma. U slučaju intracističnog krvarenja, što je neobičajena komplikacija, intenzitet signala je visok i heterogen, sa nivoom tečnosti i na T1 i na T2 snimcima, kada su prisutni mešoviti krvni produkti. Takođe, kada je sadržaj ciste bogat proteinima, može se detektovati pojačan intenzitet signala na T1 sekvenci. Softverska subtrakcija može biti korisna kada se procenjuje da li ima pojačanja signala. Mada su ovi nalazi obično precizni, pre operacije, hirurg uvek mora imati na umu mogućnost postojanja neoplastične ciste [5, 10]. Razumevanje tipičnog izgleda cističnih fokalnih lezija na jetri na snimcima skenera i magnetne rezonance omogućava jasniju dijagnozu i skraćuje vreme njenog uspostavljanja. Takođe, većina autora sugeriše sprovođenje ultrazvučne dijagnostike zajedno sa snimanjem na skeneru i magnetnoj rezonanci, kako bi se prikupilo što više informacija, i kako bi se kombinovali ovi nalazi sa kliničkim podacima, radi uspostavljanja tačne dijagnoze, pošto je od izuzetnog značaja da se pravi razlika između benignih cista i potencijalno opasnih cista, kao što su cistadenom, cistadenokarcinom i ehinokokoza, koje iziskuju specifično lečenje i terapiju [11]. U slučaju prikazanom ovde, NMR snimak pokazuje tipičan nalaz velike proste ciste u jetri, koja je iziskivala hirurško odstranjivanje.

Ciste u jetri mogu se lečiti nehirurškim ili hirurškim metodama. Perkutana aspiracija se smatra neadekvatnom, pošto je stopa ponovnog pojavljivanja ciste visoka, i postoji rizik od infekcije [12]. Saini i sar. su sprovedli studiju na 13 pacijenata sa cistama u jetri, koji su lečeni perkutanom aspiracijom iglom. Kod svih pacijenata cista se ponovo pojavila u roku od dve godine [13]. U literaturi je opisano nekoliko drugih metoda sa različitim ishodom: perkutana aspiracija sa sklerozom, terapija transdermalnim transhepatičnim injekcijama etanola, aspiracija ubrizgavanjem etanola, formalina, minociklina, monoetanolamina, doksiciklina ili oleata. Međutim, ove procedure mogu izazvati ireverzibilni sklerozirajući holangitis, zbog moguće komunikacije sa bilijarnim sistemom [5,14]. Tokom poslednjih 20 godina, otvoreni hirurški zahvati su uglavnom zamenjeni minimalno invazivnom hirurgijom. Postoji mnogo studija i serija prikaza slučajeva koje opisuju operacije velikih i masivnih cista u jetri laparoskopskim pristupom, sa stopom ponovnog pojavljivanja koja se kreće između 10% i 25% [5,15,16]. Rizik konverzije u otvorenu hirurgiju je oko 8% [5]. Većina prostih cista u jetri može se operisati laparoskopskim pristupom, ali postoje pacijenti koji iziskuju otvoreni hirurški zahvat. Studija koju su Gal i sar.

CT scan of such a cyst. Thus, the T1-weighted imaging sequence demonstrates uniform, very low signal intensity, while the T2-weighted imaging sequence shows an increased signal intensity, greater than other T2 hyperintense liver lesions, such as hemangioma. In case of intracystic hemorrhage, which is an uncommon complication, the signal intensity is high and heterogeneous, with a fluid-fluid level on both T1- and T2-weighted images, when mixed-blood products are present. Also, when the cyst content is rich in protein, an elevated T1-weighted imaging sequence signal can be detected. The use of imaging subtraction can be helpful when assessing the presence of signal enhancement. Although these findings are generally accurate, a surgeon should always bear in mind the possibility of the presence of a neoplastic cyst before surgery [5,10]. Understanding the typical CT and MR imaging appearance of cystic focal liver lesions allows a more definitive diagnosis and shortens the diagnostic work-up time. Also, the majority of authors suggest performing ultrasound imaging along with CT and MRI, for the purpose of collecting as much information as possible, and combining these findings with clinical information, in order to establish an accurate diagnosis, since it is extremely important to differentiate between benign cysts and potentially harmful cysts, such as cystadenoma, cystadenocarcinoma, and echinococcosis, which require specific treatment and therapy [11]. In the case presented here, the MRI showed a typical finding of a large simple hepatic cyst, which required surgical removal.

Liver cysts can be treated with nonsurgical methods or surgically. Percutaneous aspiration is considered inadequate, since the recurrence rate is high, and there is risk of infection [12]. Saini et al. conducted a study with 13 patients with liver cysts, which had been treated with percutaneous needle aspiration. All patients had a cyst recurrence within the space of two years [13]. Several other methods were described in literature with different outcome success: percutaneous aspiration with sclerosis, transdermal transhepatic ethanol injection therapy, aspiration with injection of alcohol, formalin, minocycline, monoethanolamine, doxycycline or oleate. However, these procedures may cause irreversible sclerosing cholangitis, because of the possible communication with the biliary system [5,14]. In the past 20 years, open surgical procedures have mainly been replaced with minimally invasive surgery. There are many studies and case report series which describe large and massive hepatic cysts being operated on with the laparoscopic approach, with the recurrence rate varying from 10% to 25% [5,15,16]. The risk of conversion to open surgery is about 8% [5]. A majority of simple liver cysts can be managed with the laparoscopic approach, but there are patients who

sproveli na 102 pacijenta sa džinovskim cistama u jetri, koji su operisani pomoću različitih tehnika, pokazala je da ne postoji razlika u kvalitetu života u zavisnosti od vrste hirurškog zahvata (minimalno invazivna hirurgija naspram otvorene) [6]. Naša pacijentkinja je operisana laparoskopski i nije pokazivala znake ponovnog pojavljivanja ciste na prvom kontrolnom pregledu. Mnogi autori zaključuju da je otvorena hirurgija prvi izbor, pogotovo ako postoji sumnja na malignitet, ili ako je cista locirana posteriorno ili u blizini velikih krvnih sudova u jetri. Međutim, pošto ne postoje randomizovane kliničke studije ili studije sa velikim brojem pacijenata u kojima se porede minimalno invazivni i otvoreni hirurški pristupi, preporučujemo personalizovani medicinski pristup, nakon analize karakteristika i same ciste ali i pacijenta.

Imajući u vidu reči čuvenog lekara, ser Vilijema Oslera, koji je smatrao da je sva veština medicine u opservaciji, ali i u svetlu ovde prikazanog slučaja, zalažemo se za personalizovani pristup kod svakog pacijenta, uz kombinovanje ključnih kliničkih informacija sa radiološkim nalazima, kako bi se uspostavila precizna dijagnoza.

Sukob interesa: Nije prijavljen.

require the open surgical approach. A study by Gall et al. conducted on 102 patients with giant liver cysts, who were operated on with different techniques, showed that there was no difference in the quality of life with regards to the type of surgery (minimally invasive vs. open surgery) [6]. Our patient was operated on laparoscopically and demonstrated no signs of recurrence on first follow-up. Many authors conclude that open surgery is the treatment of choice, especially if there is a suspicion of malignancy, or if the cyst is located posteriorly or in the proximity of large hepatic blood vessels. However, since there are no randomized clinical trials, or trials with a large number of patients comparing minimally invasive and open surgery approaches, we recommend a personalized medical approach upon analyzing the characteristics of both the cyst and the patient.

Bearing in mind the words of the famous physician, Sir William Osler, who said that the whole art of medicine is in observation, and in the light of the presented case, we advocate a personalized approach with every patient, combining crucial clinical information with radiological findings in order to establish an accurate diagnosis

Conflict of interest: None declared.

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