Surgical approach for patients with unusually located hydatid cyst

Ann. Ital. Chir., 2014 85: 50-55
pii: S0003469X13021337

Irfan Eser*, Hamza Karabag**, Samil Gunay***, Ahmet Seker****, Muazzez Cevik°, Zafer Hasan Ali Sak°°, Funda Yalcin°°°, Mehmet Salih Aydin^

Harran University, Sanliurfa, Turkey
**Medical Faculty, Department of Thoracic Surgery*
***Medical Faculty ,Department of Neurosurgery*
****Ortadogu OSM Hospital Department of Thoracic Surgery*
*****Medical Faculty Department of General Surgery*
°Medical Faculty, Department of Pediatric Surgery
°°Medical Faculty Department of Chest Diseases
°°°Medical Faculty Department of Chest Diseases
^Medical Faculty, Department of Cardio Vascular Surgery

Surgical approach for patients with unusually located hydatid cyst

INTRODUCTION: *Hydatid cyst is a parasitic disease caused by Echinococcus granulosus whose people is the intermediate host. Although this parasite can settle in any part of the human body, it is frequently seen in liver and lungs. The rate of unusual located hydatid cyst outside of liver and lungs is 13,9%. In this study, we aimed presenting unusual located hydatic cysts regarding 51 patients.*
MATERIAL AND METHOD: In *this retrospective study, the files of the patients operated in our department between 2005 and 2012 with the diagnosis of hydatid cyst, characterized be an additional location besides liver and lung involvement and located outside of liver and lung were controlled.*
FINDINGS: We *had a total of 51 patients between the ages of 6-79 (average age 35,34), 20 of them were men (39%) and the others were women (61%) (men/women = 1.56). The cysts outside of liver and lung were frequently seen in spleen (24/51), ovarium (9/51), intraabdominal (8/51), brain (8/51), kidney (6/51), psoas muscle (1/51), bladder (1/51), cervical lymph node (1/51), the heart(1/51) respectively. The most frequent symptom in our patients was stomachache. Besides, symptoms of cough, fever, respiratory disorder were present; only one patient suffered from hemophtysis. While 32 patient out of 51 were treated by laparotomy, 8 patients were operated with laparotomy and thoracotomy in the same session; the patient with 2 ovarian cysts was submitted to cystectomy through laparoscopic surgery. As a patient had a cyst both in brain and liver, he was submutted to laparotomy and craniotomy. 46 cysts in 9 patients with lung involvement were treated with lung resections: 7 wedges resection and 2 segmentectomies. The other lung cysts of the analysed patients were treated by cystectomy and capitonnage. Bile leakage was detected in a total of 7 patients: 3 of them were treated with T tube drainage and the others were endoscopically healed by means of ERCP.*
CONCLUSION: *The incidence of hydatid cyst, which is an important health problem in endemic areas, can be reduced by means of simple preventive measures. Its basic treatment is surgery. Main objective of the surgery should be parenchyma sparing while taking off completely the cysts. Although the disease is frequently seen in liver and lung, other organ involvements should be considered. Thus, it does not matter where hydatid cyst is seen, abdomen and thorax should be attentively controlled by the simplest imaging method also outside of clinical symptoms. Abdomen and thorax imaging should be carried out at least once in the two following years to have an early detection of an eventual recurrence.*

KEY WORDS: Hydatid Cyst, Parenchyma Preservation Surgery, Unusual hydatid cyst localization

Pervenuto in Redazione Febbraio 2013. Accettato per la pubblicazione Aprile 2013
Correspondece to: Irfan Eser, MD, Assistant Professor of Thoracic Surgery, Harran University Faculty of Medicine, Department of Thoracic Surgery Sanliurfa, Turkey (e-mail: drirfanes@gmail.com)

Introduction

Hydatid cyst is a parasitic disease caused by parasite of Echinococcus granulosus which is human's intermediate hote. It is frequent in the Southern America, Australia, New Zealand, Russia and The Mediterranean Countries where animal breeding is dense in the world. This disease in our country is seen as endemic. Even if this parasite locates anywhere in human body, the most frequent location areas are liver and lungs. The rate of the rare located hydatid cyst out of liver and lung is 13,9%. In compliance with the order of frequency, among rare location areas are spleen, soft tissue, intra-abdomen, kidneys, pancreas, breast, pelvis, articular, vesica urinaria, heart, ovarium, thyroid, retroperitoneum, incision scar and choledoch [1,2]. Heart cyst hydatic can be misdiagnosed as coronary artery disease, cardiac valvular disease and pericarditis. After surgical excision without rupture for cardiac hydatid cyst, medical therapy is performed to prevent recurrence. According to the ones notified in a study made in Australia, organs like liver (63%), lungs (25%), muscles (5%), bone (3%), kidney (2%), cardiac %0.02-2, brain (1%), spleen (1%) can be held in the disease of hydatid cyst [3,4]. We aimed at presenting unusually located hydatid cysts covering 51 patients in this study.

Material and Method

In this study, the files of the hydatid cyst diagnosed patients who were operated consecutively in our region between the dates of 2005-2012 were scanned in the archives of our hospital retrospectively. The files of the patients who has only lung or only liver located or only liver-lung located patients were left out of study.

Findings

Our patients are composed of 51 patients, 18 of them who are between 6-79 years old (average age of them is 35, 34) are male and 33 of them (66%) are female. Even if that the female number is twice more than male number is supported by literature, it may be due to 5 cysts stemmed from ovarium (Table I).

The most frequently seen symptom on patients was stomachache. Besides this, cough, fever, dyspnoea complaints were available, in only one patient there was hemoptysis. Cysts grow slowly and don't generally show indications until they reach diameter of 5 cm. As the size becomes bigger, it shows clinical findings due to pressure and occlusive effects [5,6]. If rare locations aren't kept in mind, cyst rupture, fever, eosinophilia, findings going to anaphylactic shock and complications like suppuration can occur. For all the patients, according to location points, the sophisticated methods like USG, MR and tomography were used. Stomach resection was applied to the patient who had a cyst nearly 12 cm stuck to stomach wall in abdomen. Splenectomy was applied to 8 of 24 patients who have spleen involvement. The patient who has bilateral lung, right atrium, liver and spleen involvement was followed by medical treatment accepted as inoperable (Fig. 1).

TABLE I - *Unusual settlement locations and the number of patients.*

Localization	Number of patients Men	Women	Total
Liver+ Spleen	3	8	**11**
Brain	4	4	**8**
Spleen	4	1	**5**
Liver+ Ovary		5	**5**
Liver + Omentum	2	3	**5**
Lung + Spleen	3		**3**
Lung + Kidney		2	**2**
Liver + Kidney		2	**2**
Liver +Spleen+ Kidney	1		**1**
Lung+ Liver + Spleen+Omentum		1	**1**
Lung+Liver+ Spleen		1	**1**
Lung+Liver+Omentum		1	**1**
Liver + Brain	1		**1**
Omentum + Uriner Bladder		1	**1**
Servical lymph node	1		**1**
Kidney+ omentum		1	**1**
Liver + Psoas muscle		1	**1**
Lung+Liver+ Spleen+heart	1		**1**
Total	**20(%34)**	**31(%66)**	**51**

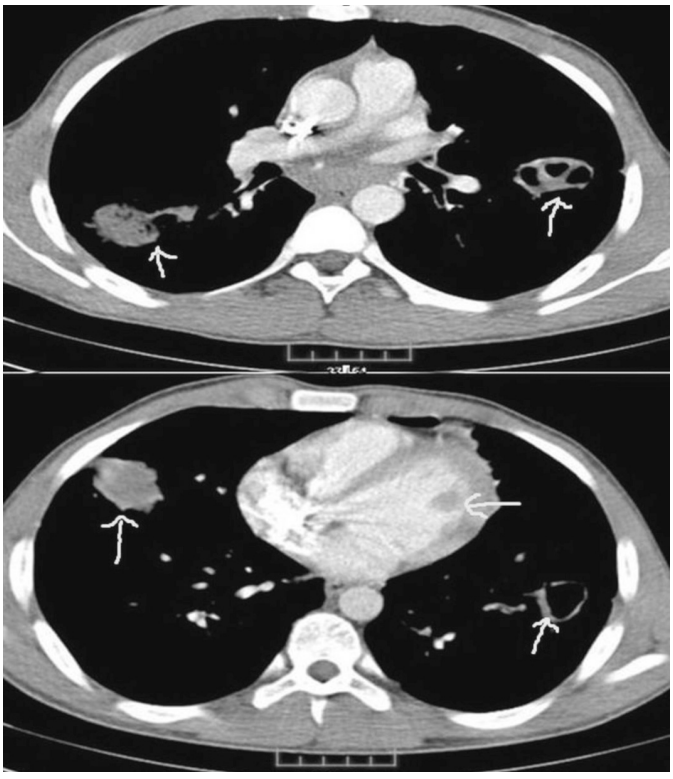

Fig. 1: Bilateral Lung and Heart.

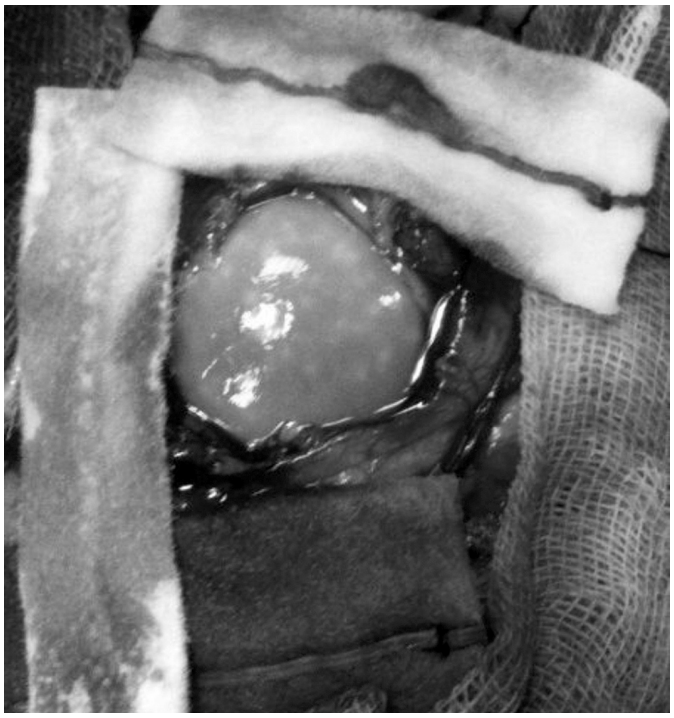

Fig. 2: Intracranial cyst enucleation.

Fig. 3: Removed intracranial cyst.

TABLE II - *The organ involving cyst, total cyst and number of patients.*

The organ involving cyst	Number of cyst	Number of patients
Liver	46	27
Spleen	25	24
Lung	25	9
Ovary	11	9
Omentum	8	8
Brain	8	8
Kidney	6	6
Psoas Muscle	1	1
Urinar Bladder	1	1
Servical Lymph Node	1	1
Heart	1	1
Total	**133**	

TABLE III - *Access methods applied*

Access methods applied	
Laparotomy	32
Thoracotomy+ Laparotomy	8
Craniotomy	7
Laparoscopy	2
Laparotomy + Craniotomy	1
neck lymph node dissection	1
Total	51

TABLE IV - *Operations applied on cyst*

Operations pplied on cyst	
Cystectomy	78
cystotomy+ Capitonnage	37
Splenectomy	7
Wedge resection	7
Segmentectomy	2
Salpingectomy	1
Gastric resection	1
Total	133

In the patient who came after the traffic accident and where the cyst was detected in the examinations made, there were 8 cysts in left lower lobe, 5 in left upper lobe and 1 in spleen. Segmentectomy were applied to left lower lobe in lungs, cystotomy and capitonnage were applied to cysts left behind. In the same session for spleen splenectomy was applied after laparotomy. In the follow-up after the operation air leak in lungs and bleeding inside abdomen were determined. After relaparotomy, in the postop period the patient died. In 8 patients, intracranial hydatid cyst was determined and operated. Two of them died after the operation (Figg 2 and 3). 133 cysts were determined in 51 patients totally. The number of cyst per each patient was 2,6. The most cyst (46 cysts) was seen in liver. In one patient unity of liver, lung, spleen and heart was determined and this patient was applied only medical treatment (Table II). While 32 of 51 patients were applied laparotomy, both laparotomy and thoracotomy were applied to 8 patients in the same session; cystectomy was applied to the

patient who has two ovarian cysts by laparoscopic surgery. In one patient since there is one cyst in both brain and liver, laparotomy and craniotomy were applied (Table III). 7 wedge and two segmentectomies were applied to 46 cysts in 9 patients that show involvement in lungs. Cystotomy and capitonnage were applied to the cysts of lungs left (Table IV). Bile leakage was determined in 7 patients totally. T Tube drainage was applied to three of them and ERCP to four.

Discussion

Hydatid cyst disease has been known since Galen and Hippocrate's time. The disease was first described by Thebesius in 17th C and was named as hydatid cyst by Rudolphi in 1808 [7,8]. Especially, it is a parasitic disease seen frequently in the countries where preventive medicine isn't enough and which deal with the animal husbandry. While the disease occurs on liver and lungs, it is also seen on whole body. Hydatid cyst disease can occur on liver (63%), lungs (25%), muscles (5%), bone (3%), kidney (2%), brain (1%), spleen (1%) [9]. Our study is quite rare in terms of the number of case for unusual locations of the hydatid cyst disease. In this study, we saw that there was spleen involvement more than notified in literature. Besides this, while the soft tissue involvement which is seen quite frequently in unusual locations is seen rarely, bone and skin involvement wasn't seen.

In childhood, lung involvement is seen more frequently than liver involvement. Most frequently, right lung and lower lobe of right lung is caught [10]. While primer hydatid cysts of peritoneal cavity are seen rarely, secondary multiple cysts are seen more frequently. Most of the secondary cysts develop due to the rupture of the cyst in liver [11]. Ethiopathogenesis in secondary originated diffused abdominal hydatidosis is related to the fact that fertile scolexes are implanted in periton after the main hydatid cyst in liver is torn. 10% of the hydatid cysts of liver ruptures peritoneal cavity. Even if these ruptures follow abdominal trauma, sometimes it can occur spontaneously. Even if the first treatment option is considered as surgery, the condition is followed by medical treatment when there are quite a lot of cysts in an organ or more than one cyst in more than one organ. The aim of surgical treatment is to remove the cyst protecting the organ tissue in maximum and close the space left. The choice of surgical technique can change according to the pre-operation and post-operation findings, the experience of the surgeon and choice [12-14]. Radical surgical procedure for liver hydatidosis must be the treatment of choice rarely leading to severe and disabling complications and without risks and relapses. The choice of type of surgery must be done according to anatomic and clinic tools and experience and agreement of surgical team [15]. surgical procedures for liver hydatic cysts should be performed to each patient avoid high surgical risk due to the benign nature of the disease [16].

Bilateral lung location in one of our patients (1 on right, 3 on left), 2 right atrium on liver and the patient who has cyst location on spleen were followed medically. In medical treatment if Albendazol is administered as 3 cycle for 28 days, the rate of killing optimal parasite is achieved [17-18]. But, after albendazol is used on the soft tissues especially like lungs it is said that it has made the wall of cyst thin and then cysts have become rupture and spread. As a result of the blowing up of the cysts before they become noninfectious especially in people that the drug use isn't proper, it is known that the cysts which have blown up locate in new spaces in lungs which don't have a natural tamp like liver or spleen. In hydatid cyst disease, in involvement of multi organs as the result of surgical treatment, mortality and morbidity rates change according to the organ involved and the number of organ involved and the number of cyst. But, as the resection of the organ which is involved rarely in unusual locations is much more, the rates of recurrence are low. In our series only in two cases recurrence seemed. One of the recurrences was liver located and the other was intra-abdominal [19].

In our series in surgical approach to cysts, the procedure protecting parenchyma was applied. In our series of 51 patients the rate of mortality is determined as 5.8% (3/51). In our study series among the rarely seen locations, involvement of lymph node on neck (superficial servical lymph node) was seen, too. Convulsion and fainting was available on three of our 8 patients who showed intracerebral location.

Since our region is an endemic area in regard of hydatid cyst, even in findings which remind the hydatid cyst doctors in the region think about the hydatid cyst disease. Nevertheless; to bump into a patient who has such quite a lot of number of organ involvement can be explained like that generally patients don't apply to a health center before they are caught the disease completely. It isn't hard to diagnose hydatid cyst with a careful anamnesis and physical examination when the clinician thinks of. Although the diagnosis before the operation becomes easier by the developments in imaging methods, in order that the cyst can be distinguished from tumor, abscess or the other simple cysts which take up space, and recurrences are evaluated truly, it must be supported by serologic diagnosis methods. For that purpose, Indirect Hemagglutination Antibody Test and Latex Agglutination Test are the tests used most frequently. In addition, ultrasonography, computerized tomography and magnetic resonance imaging techniques are so sensitive and not invasive.

Conclusion

The frequency of hydatid cyst which is a great problem in endemic areas can be diminished by simple preventing

cautions. Its basic treatment is surgery. The main goal in surgery must be parenchyme protecting. Our study has shown us that even if the unusual located hydatid cysts are the disease which can be seen on every part of the body with multi organ involvement, the mortality and morbidity of which are high, a good physical examination and a simple radiological screening by anamnesis can diagnose the disease comfortably. Even if the disease locates frequently on liver and lungs, the other organ involvements should be kept in mind. Therefore, wherever the hydatid cyst is seen on the body abdomen and thorax must be screened completely by the simplest screening method which can be reached. In the follow-ups recurrence must be evaluated screening abdomen and thorax in the first two years twice in a year absolutely.

Riassunto

La cisti idatidea è una malattia parassitaria causata dall' Echinococcus granulosus di cui l'uomo è l'ospite intermedio. Nonostante possa localizzarsi in ogni parte del corpo umano essa viene più frequentemente rinvenuta nel fegato e nei polmoni. La frequenza delle localizzazioni al di fuori di fegato e polmoni è del 13,9%. In questo studio ci siamo proposti di presentare la localizzazione rara della cisti idatidea in 51 pazienti.
Si tratta di uno studio retrospettivo, basato sul controllo delle cartelle cliniche dei pazienti operati nel nostro dipartimento tra il 2005 ed il 2012 con la diagnosi di cisti idatidea, caratterizzati da localizzazioni aggiuntive rispetto a fegato e polmoni comunque interessati.
L'età dei 51 pazienti risulta compresa tra 6 e 79 anni (media 35,34), di cui 20 uomini (39%) ed il restante donne (61%) con un rapporto uomo/donna = 1,56). La localizzazione della cisti al di là del fegato e del polmone è stata rispettivamente nella milza (24/51), nell'ovaio (9/51), nella cavità addominale (8/51), nel cervello (8/51), nel rene (6/51), nel muscolo psoas (1/51), nella vescica (1/51), in un linfonodo cervicale (1/51) e nel cuore (1/51). La sintomatologia più frequente nei nostri pazienti è stata la gastralgia. Inoltre erano presenti tosse, febbre e disturbi respiratori; soltanto un paziente ha manifestato emottisi.
Mentre 32 pazienti sono stati trattati con una laparotomia, 8 sono stati operati con laparotomia e toracotomia nella stessa seduta; la paziente con due cisti dell'ovaio fu trattata con cistectomia per via laparoscopica. Il paziente con cisti nel fegato e nel cervello è stato trattato con laparotomia e craniotomia.
46 cisti idatidee in 9 pazienti con localizzazione polmonare sono state trattate con resezione polmonare: 7 resezioni cuneiformi e due segmentectomie. Le altre cisti polmonari sono state trattate con cistectomia e capitonnage. Fistole biliari si sono avute in un totale di 7 pazienti, 3 dei quali trattati con drenaggio secondo Kehr e gli altri per via endoscopica mediante ERCP.
In conclusione l'incidenza delle cisti idatidee, che rappresenta un importante problema sanitario nelle aree endemiche, può essere ridotta con semplici misure preventive. Il loro trattamento di base è la chirurgia il cui principale obiettivo deve essere quello conservativo nei confronti del parenchima pur realizzando l'asportazione completa delle cisti.
Sebbene la malattia è di frequente osservata a livello del fegato e dei polmoni bisogna tenere presente la possibilità di coinvolgimento di altri organi. Pertanto non è importante ove la cisti viene diagnosticata, ma sia l'addome che il torace devono essere controllati attivamente con il più semplice dei mezzi di imaging anche al di fuori della sintomatologia clinica. L'imaging dell'addome e del torace dovrebbero essere ripetute almeno entro i due anni successivi al fine di una diagnosi precoce delle recidive.

References

1. Kiresi DA, Karabacakoglu A, Odev K, Karakose S: *Uncommon locations of hydatid cysts*. Acta Radiol, 2003; 44:622-36.

2. Hakverdi S, Sayar H, Yaldiz M, Erdogan S, Akansu B, Canda MS: *Cukurova yoresinde seyrek yerlesimli ekinokokkozis.* Türk Parazitol Derg, 2009; 33:77-81.

3. Amman R: *Echinococcus.* Gastroenterol Clin N Am, 1996; 25: 655-89.

4. Kurdal AT, Kahraman N, Iskesen I, Sirin BH: *Unusual location of hydatid cyst: The posterior leaflet of tricuspid valve*. Ann Ital Chir, 2010; 81(3):211-14.

5. Altintas N, Tinar R, Çoker A: *Echinococcosis.* Baski In: Altintas N, Tinar R, Çoker A(eds); Hidatoloji Dernegi Yayin, 2004; 1:129-283.

6. Czermak BV, Akhan O, Hiemetzberger R, Zelger B, Vogel W et al: *Echinococcosis of the liver.* Abdominal Imaging, 2008; 33(2): 133-43.

7. Varela A, Burgos R, Castedo E: *Parasitic diseases of the lung and pleura*. In: Patterson GA, Cooper JD, Deslauriers J (eds): *Pearson's thoracic & esophageal surgery.* 3rd ed. Philadelphia: Churchill Livingstone, 2008; 550-65.

8. Harlaftis NN, Aletras HA, Symbas PN: *Hydatid disease of the lung*. In: Shields TW, Locicero III J, Reed CE, Feins RH (eds) *General thoracic surgery*. 7th ed. Philadelphia:Lippincott Williams & Wilkins,2009; 1187-195.

9. Amman R: *Echinococcus.* Gastroenterol Clin N Am, 1996; 25: 655-89.

10. Kurul IC, Topcu S, Altinok T, Yazici U, Tastepe I, Kaya S, et al.: *One-stage operation for hydatid disease of lung and liver: Principles of treatment*. J Thorac Cardiovasc Surg, 2002; 124:1212-215-512.

11. Demirel AH, Akgün A, Öngören AU, Kisakürek M, Erol MF: *Atipik lokalizasyonlu kist hidatikler hydatid cyst cases with atypical location*. Akademik Gastroenteroloji Dergisi, 2007; 6 (3): 158-60 Olgu Sunumu.

12. Schantz PM: *Echinococcus Species (Agents of Cystic, Alveolar, and Polycystic Echinococcosis)*. In: Long SS, Pickering LK, Prober CG. (Eds): *Principles and practice of pediatric infectious diseases.* Second edit. New York; Churchill-Livingstone 2003; 1357-356.

13. Sbihi Y, Rmiqui A, Rodriguez-Cabezas MN, Orduña A, Rodriguez-Torres A, Osuna A: *Comparative sensitivity of six serological tests and diagnostic value of ELISA using purified antigen in hydatidosis*. J Clin Lab Anal, 2001; 15: 14-18.

14. Gonlugur U, Ozcelik S, Gonlugur TE, Celiksoz A: *The role of Casoni's skin test and indirect haemagglutination test in the diagnosis of hydatid disease*. Parasitol Res, 2005; 97:3959-8.

15. Sciumè C, Geraci G, Pisello F, Facella T, Li Volsi F, Modica G: *Surgical treatment of liver hydatidosis: Our experience with diagnostic and therapeutic consideration*. Ann Ital Chir, 2005; 76(2):147-53; discussion 153-5 Italian.

16. Gruttadauria S, Basile F, Marino G, Gentile A, Vittoria Sgroi AV, Gruttadauria G: *Development in diagnosis and treatment of hepatic echinococcosis in a surgical department of a Mediterranean centre over a 20-years period*. Ann Ital Chir, 2000; 71(1):99-104; discussion 105.

17. Silva MA, Mirza DF, Bramhall SR, Mayer AD, McMaster P, Buckels JA: *Treatment of hydatid disease of the liver. Evaluation of a UK experience*. Dig Surg, 2004; 21(3):227-33; discussion 233-4. Epub 2004 Jun 30.

18. Jevtic M, Mikic D, Arsic-Komljenovic G, Stankovic N, Ristanovic E, Sjenicic G, Janicijevic-Hudomal S.: *Adverse effects of longterm, continual administration of high doses of albendazole in the treatment of echinococcal disease.* Vojnosanit Pregl, 2008; 65(7):539-44. Serbian.

19. Versaci A, Scuderi G, Rosato A, Angio LG, Oliva G, et al.: *Rare localisation of echinococcosis: Personal experience*. ANZ J Surg, 2005; 75:986-91.