



Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Difficulties in diagnosing the alveolar echinococcosis (case report)

Dinar Kaliyeva^a, Yekaterina Yukhnevich^{b,*}, Nurkassi Abatov^a, Aydin Nurbekov^a^a Department of Surgical Diseases, Karaganda Medical University, 40, Gogolia str, Karaganda 10017, Kazakhstan^b Department of Clinical Pharmacology and Evidence-Based Medicine, Karaganda Medical University, 40, Gogolia str, Karaganda 10017, Kazakhstan

ARTICLE INFO

Article history:

Received 7 June 2020

Received in revised form 24 August 2020

Accepted 24 August 2020

Available online 31 August 2020

Keywords:

Alveolar echinococcosis

Large size

Differential diagnosis

ABSTRACT

INTRODUCTION: Alveolar echinococcosis is dangerous parasitic zoonose with the large endemic area. This disease has a high prevalence in Kazakhstan.**PRESENTATION OF CASE:** We report on a 45-year woman suffering from alveolar echinococcosis with a huge cystic mass and difficulty of differential diagnosis. She was hospitalized for surgery with primary diagnosis of hydatid disease. The liver carcinoma was suspected during surgery due to the huge size and structure of the mass. This mass was totally removed. The alveolar echinococcosis was confirmed by histopathological examination.**DISCUSSION:** Essential features of this case are the large size of the lesion with a dense consistency and the germination of blood vessels, which unusual for alveolar echinococcosis. Total resection and Albendazole therapy was successful for patient, she didn't have a relapse during the follow-up examination.**CONCLUSION:** This report may provide new aspects of visualization of alveolar echinococcosis and highlight the necessity for the upgrade of the diagnosis tactic.© 2020 The Authors. Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

Alveolar echinococcosis is parasitic disease associated with high mortality and morbidity [1,2] and is very common in Asia [3,4]. Alveolar echinococcosis is also widely distributed in some European countries and detected mainly in people aged 18–65 years [5,6]. The disease is usually asymptomatic for a long period of time and differential diagnosis required after symptoms appear. Alveolar echinococcosis as with hydatid disease is often expensive and complicated to treat. Most patients require surgery with prolonged drug therapy. We report a case of hepatic alveolar echinococcosis in a 45-year old female patient with a huge cystic mass which presented a difficult differential diagnosis. This work has been reported in line with the SCARE criteria [7].

1. Case presentation

A 45-year-old woman was admitted to Regional Hospital (Kazakhstan) with a history of recurrent pain in the right upper abdomen. She was examined in the outpatient clinic and ultrasonography showed a huge cystic mass in the liver. A hyperechoic lesion with heavy capsule was detected given the impression parasites in the right lobe of the liver in the projection of V-VII segments. The patient was admitted for surgical treatment of hydatid disease. Subsequent computed tomography demonstrated a large cystic lesion (Fig. 1) and the patient underwent surgical exploration.

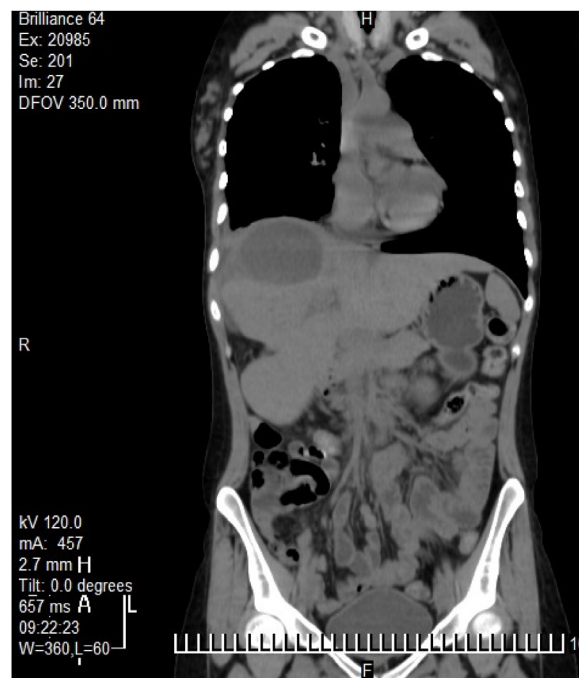


Fig. 1. Computed tomography of the liver demonstrating a cystic lesion in S_{VII}-S_{VIII} with irregular shape and size 8.3 × 6.5 cm.

* Corresponding author.

E-mail address: 3204229@gmail.com (Y. Yukhnevich).

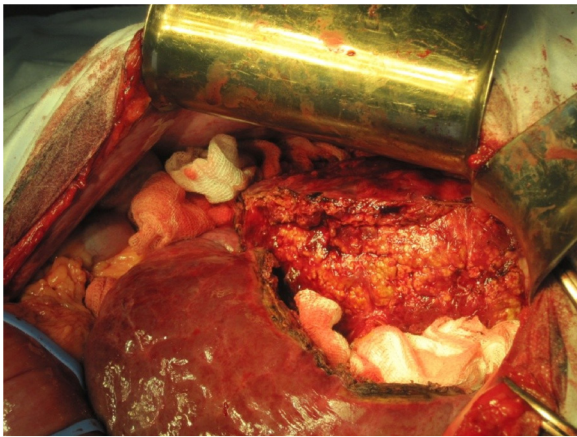


Fig. 2. Operative picture demonstrating the lesion in all hepatic right lobe.



Fig. 4. The macroscopic examination of the size of mass.



Fig. 3. Last stage of cyst isolation.

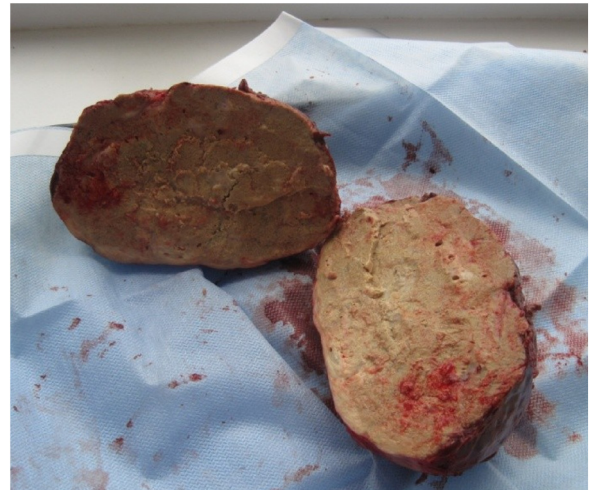


Fig. 5. The internal contents of mass.

Intraoperative exploration revealed a massive formation with stone-like density occupying the whole right lobe of the liver (Fig. 2). It was assumed that the patient had liver carcinoma and therefore the enucleation of this huge mass was performed with maximum protection of the remaining liver tissue. The separation of mass from liver tissue was conducted in stages with gradual vascular ligation. A dense mass spread across all the depths of liver tissue. There was a clear boundary between liver tissues and the lesion. The extraction of the lesion was performed after phasing separation (Fig. 3).

Examination of the remaining part of the liver showed that only small part of the left liver lobe was undamaged by the cyst. Due to the scale of the liver injury peritoneal repair with omentum was performed. The drainage tubes were left in subphrenic space and pelvic area to prevent postoperative complications.

Examination after surgery showed a mass of dense consistency with large numbers of sprouted vessels supplying it with blood. The size of mass was $14.0 \times 11.0 \times 16.0$ cm (Fig. 4).

On dissection of the mass the internal contents were grey in color with multiple lumens of blood vessels and biliary ducts (Fig. 5). The provisional diagnosis was liver carcinoma, but this was changed when pathological examination revealed hepatic alveolar echinococcosis.

The postoperative period of the patient proceeded favorably. She received antibacterial therapy and fluid maintenance. The drainage tubes were removed on day 7 and the sutures of postoperative wound was removed on day 10 after surgery. The anti-infective drug treatment with albendazole was continued

long-term. At one year follow-up, there were no signs of cyst recurrence.

2. Discussion

Alveolar echinococcosis is a severe and potentially fatal helminthic disease with increasing incidence in endemic regions [8]. This disease is often diagnosed at an advanced stage due to lack of early symptoms and surgical treatment with complete resection is only achievable in the minority of patients [9].

This case demonstrated the challenges involved in the diagnosis of alveolar echinococcosis in that the diagnosis was not identified correctly both at initial presentation and at the intraoperative stage. Before surgery a liver cyst was suspected, taking into account the clinical and scan examination. However, during surgery it was found that the lesion was occupying the whole right lobe of the liver with a dense consistency and the proliferation of blood vessels. The resection of the right lobe was performed with suspected liver carcinoma. Finally, hepatic alveolar echinococcosis was confirmed by histopathological examination of biopsy material obtained during the surgery. Unfortunately, the serological investigation for alveolar echinococcosis confirmation is not widely available in Kazakhstan.

Examples of the problem of alveolar echinococcosis diagnosis have been previously reported in the literature. Most of them describe extrahepatic locations of the parasite which had metasta-

sized to the brain and lungs or difficulty in differentiating alveolar echinococcosis from liver cancer because of the invasion of biliary and vascular tissue of the liver. In this case a patient undergoing an operation by operated by neurosurgeons for suspected metastatic cancer of the liver and brain is described. A alveolar echinococcosis of the liver and brain was verified by the histological examination of the biopsy [10]. In a further case a patient presented with symptoms of acute cholecystopancreatitis. An atypical primary liver cancer was suspected during examination and an interventional biopsy of the lesion was preoperatively performed. The histological findings were consistent with alveolar echinococcosis, but the macroscopic pattern observed during surgery was typical for cystic echinococcosis [11]. There are also cases of extrahepatic alveolar echinococcosis without liver involvement, when the involvement of the spleen and lungs or only the spine was observed [12].

The diagnostic difficulties presented in these publications indicate the importance of the problem under consideration.

In conclusion, these cases emphasize the problem associated with difficulties with diagnosis of alveolar echinococcosis.

Declaration of Competing Interest

The authors report no declarations of interest.

Sources of funding

None. The surgery was performed in government hospital and was free.

Ethical

None. There were not research interventions, all therapy was according to national guidelines.

Consent

The signed consent to publish a case report from the female patient was received.

Author's contribution

Kaliyeva D. was surgery assistant in operation, she presented the case report and gave the description and analysis of case.

Yukhnevich Y. provided the collection of literature data, took part in analysis and performed the translation in English, she is also corresponding author.

Abatov N. is foremost surgeon in this difficult surgery, he gave suggestion to publish this case, participated in the concept and revised of the manuscript.

Nurbekov A. was surgery assistant in operation, participated in the concept and revised of the manuscript.

Registration of Research Studies

None.

Guarantor

Yekaterina Yukhnevich.

Provenance and peer review

Not commissioned, externally peer-reviewed.

References

- [1] P.R. Torgerson, K. Keller, M. Magnotta, N. Ragland, The global burden of alveolar echinococcosis, *PLoS Negl. Trop. Dis.* 4 (2010), <http://dx.doi.org/10.1371/journal.pntd.0000722>.
- [2] P. Deplazes, L. Rinaldi, C.A. Alvarez Rojas, P.R. Torgerson, M.F. Harandi, T. Romig, et al., Global distribution of alveolar and cystic echinococcosis, *Adv. Parasitol.* 95 (2017) 315–493, <http://dx.doi.org/10.1016/bs.apar.2016.11.001>.
- [3] S. Baumann, R. Shi, W. Liu, H. Bao, J. Schmidberger, W. Kratzer, et al., Worldwide literature on epidemiology of human alveolar echinococcosis: a systematic review of research published in the twenty-first century, *Infection* 47 (2019) 703–727, <http://dx.doi.org/10.1007/s15010-019-01325-2>.
- [4] H. Wen, L. Vuitton, T. Tuxun, J. Li, D.A. Vuitton, W. Zhang, et al., Echinococcosis: advances in the 21st century, *Clin. Microbiol. Rev.* 32 (2019), <http://dx.doi.org/10.1128/CMR.00075-18>.
- [5] J. Fabbri, C.M. Clemente, N. Elisondo, G. Gambino, S. Ravetti, L.Y. Hergert, et al., Anti-echinococcal activity of menthol and a novel prodrug, menthol-pentanol, against *Echinococcus multilocularis*, *Acta Trop.* 205 (2020), <http://dx.doi.org/10.1016/j.actatropica.2020.105411>.
- [6] G. Nunnari, M.R. Pinzone, S. Gruttadauria, B.M. Celesia, G. Madeddu, G. Malaguarnera, et al., Hepatic echinococcosis: clinical and therapeutic aspects, *World J. Gastroenterol.* 18 (2012) 1448–1458, <http://dx.doi.org/10.3748/wjg.v18.i13.1448>.
- [7] R.A. Agha, M.R. Borrelli, R. Farwana, K. Koshy, A. Fowler, D.P. Orgill, For the SCARE Group, The SCARE 2018 statement: updating consensus Surgical Case Report (SCARE) guidelines, *Int. J. Surg.* 60 (2018) 132–136.
- [8] L.A. Salm, A. Lachenmayer, S.F. Perrodin, D. Candinas, G. Beldi, Surgical treatment strategies for hepatic alveolar echinococcosis, *Food Waterborne Parasitol.* 15 (2019), e00050, <http://dx.doi.org/10.1016/j.fawpar.2019.e00050>.
- [9] J. Schmidberger, J. Steinbach, P. Schlingeloff, W. Kratzer, B. Grüner, Surgery versus conservative drug therapy in alveolar echinococcosis patients in Germany – a health-related quality of life comparison, *Food Waterborne Parasitol.* 16 (2019), e00057, <http://dx.doi.org/10.1016/j.fawpar.2019.e00057>.
- [10] D.P. McManus, Z. Li, S. Yang, D.J. Gray, Y. Yang, Case studies emphasising the difficulties in the diagnosis and management of alveolar echinococcosis in rural China, *Parasites Vectors* 4 (2011), <http://dx.doi.org/10.1186/1756-3305-4-196>.
- [11] G. Atanasov, C. Benckert, A. Thelen, D. Tappe, M. Frosch, D. Teichmann, et al., Alveolar echinococcosis-spreading disease challenging clinicians: a case report and literature review, *World J. Gastroenterol.* 19 (2013) 4257–4261, <http://dx.doi.org/10.3748/wjg.v19.i26.4257>.
- [12] S. Reuter, H.M. Seitz, P. Kern, T. Junghanss, Extrahepatic alveolar echinococcosis without liver involvement: a rare manifestation, *Infection* 28 (2000) 187–192, <http://dx.doi.org/10.1007/s150100050079>.

Open Access

This article is published Open Access at [sciencedirect.com](https://www.sciencedirect.com). It is distributed under the [IJSCR Supplemental terms and conditions](#), which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.