Case Report

A rare presentation of hydatid cyst, involvement of uncommon sites with sparing of typical locations

Mehdi Karami[1], Seyedeh Fatemeh Sadatmadani[1], Hossein Kouhi[1], Bahar Sadeghi[1], Zeynab Rostamiyan[1], Mozhdeh Hashemzadeh[2]

[1]Department of Radiology, School of Medicine, Isfahan University of Medical Sciences, Isfahan, Iran, [2]Clinical Informationist Research Group, Health Information Research Center, Isfahan University of Medical Sciences, Isfahan, Iran

Hydatid cyst (HC) is a serious health problem in endemic areas. Liver and lung are the most common involved organs while the involvement of muscles, heart, kidney, brain, and spleen is rare. The involvement of these rare locations for HC is mostly in association with infestation of common sites. We report a case of 43-year-old man with a history of surgery of HC in brain and heart who complained from chest pain. In imaging modalities, three cystic lesions were seen in heart and kidney which were confirmed to be recurrence of HC by serologic tests. However, no evidence of involvement of liver and lungs as the most common infected organs of HC was seen in recent and previous imaging mo°dalities. The patient refused another surgery and just accepted anthelmintic drugs. The practical point of this case report is that when we see any cystic lesion in imaging modalities in uncommon sites for HC, while common sites are spare, we should still consider the possibility of HC and not rule out it just because of lack of simultaneous infestation of its common involved organs.

Key words: Echinococcosis, hydatid cyst, hydatid disease

How to cite this article: Karami M, Sadatmadani SF, Kouhi H, Sadeghi B, Rostamiyan Z, Hashemzadeh M. A rare presentation of hydatid cyst, involvement of uncommon sites with sparing of typical locations. J Res Med Sci 2021;26:1.

INTRODUCTION

Human hydatid cyst (HC) or cystic echinococcosis is a life-threatening neglected zoonotic infectious disease that occurs in both developing and developed countries, and is recognized as a serious public health problem.[1,2]

The most common affected organs in HC are liver and lungs where 90% of the echinococcal cysts develop and 10% of cysts are in anywhere of the body such as brain, Heart and kidney which are considered rare sites of HC localization.[3] Infestation of rare sites is usually in correlation with common organs involvement. HC can lead to severe complications with morbidity, mortality and poor prognosis. HC diagnosis is very important and is based on clinical history, physical examination, a series of serologic tests and imaging findings.[2,4] We report a case presented with HCs in brain, heart and kidney as rare organs of HC involvement; while the most common infected organs such as liver and lungs were intact. Results of search in BMJ, PubMed, ISI, and Google scholar reported 15 similar cases. However, the cases were not found to be quite similar to the present case.

CASE REPORT

Our patient is a 43-year-old man who 3 years' ago complained from headache. In brain magnetic resonance imaging (MRI), HC (16 mm × 22 mm) in the left parieto-temporal lobe was found [Figure 1] and he underwent brain surgery. After brain surgery antihelminthic conservative treatment was performed. One year later, he returned to hospital with chest pain and in investigations cardiac HC in posterior wall of the left ventricle (LV) with dimensions of 17 mm ×

Access this article online

Quick Response Code:

Website: www.jmsjournal.net

DOI: 10.4103/jrms.JRMS_127_20

Address for correspondence: Dr. Hossein Kouhi, Department of Radiology, School of Medicine, Isfahan University of Medical Sciences, Isfahan, Iran. E-mail: hkoohi91@gmail.com

Submitted: 04-Feb-2020; **Revised:** 15-Mar-2020; **Accepted:** 18-Aug-2020; **Published:** 28-Jan-2021

21 mm was found and cardiac surgery was done for his treatment. From a few weeks before his recent referral (to Chamran Hospital of Isfahan, Iran), he complained of localized burning pain in his left hemi-thorax and left arm. He expressed a history of fever, chills, cough, and sputum from some days ago.

Important laboratory test findings were: hemoglobin: 14 g/dl (normal range = 14–18 g/dl), erythrocyte sedimentation rate: 6 mm/h (normal range = 0–20 mm/h), troponin in normal range (<0/04 ng/dl), wright 2ME: negative, immunoglobulin E: 332 IU/mL (normal range = <2000 IU/ml, white blood cells: 5700 UL/10 × 3 (normal range = 3.5–11ul/10 × 3). Abdominopelvic ultrasound showed a cystic lesion in the upper-middle zone of the left kidney containing a septum in diameter of 25 mm. Electrocardiogram showed the right bundle branch block. Chest and abdominal MDCT revealed an intramyocardial well-defined round cystic mass (16 mm × 22 mm) in posterolateral aspect of LV and one mass (37 mm × 23 mm) in pericardium and also showed the left renal cyst [Figures 2 and 3]. The patient refused surgery and was discharged with personal consent. Oral albendazole (400 mg twice a day) was prescribed for him. After being discharged from hospital, because the patient stopped cooperating with the treatment team, we could not follow-up with him.

DISCUSSION

HC is a zoonotic and endemic infection in some areas, such as South America, North Africa, Asia, and Australia. Humans become infected with the parasite (*Echinococcus granulosus*, *Echinococcus multilocularis*) by ingesting embryonated eggs shed in canids' faeces. Embryos are released in small gut and migrate to the organs through the blood-stream. Hepatopulmonary HCs are common in hydatid infection; because liver is the first and the most frequent involved organ (70% cases) and the second common infected site is lung (20%).[2-4] An interesting point of our case was that typical organs for HC were intact and other sites were infected.

Uncommon affected organs and their prevalence in HC include muscle (5%), heart (0.5%–2%), kidney (2%), brain (1%), and spleen (1%). Furthermore, extra hepatopulmonary HCs are reported at orbit, parotid, thyroid, gallbladder, adrenal, mesentery, pancreas, ovary, and tibia. In 20%–40% of cases, like our case, multiple cysts or organs involvement are reported. Involvement of rare locations is usually in association with concurrent common sites infestation, such as after cyst rupture. However, in our case, this correlation was not seen.[2-11]

As it mentioned, cardiac, cerebral and renal HC involvement are atypical and uncommon that our case had those. The most common involved part in cardiac HC is LV because of a rich coronary blood supply. Our case had two cysts in heart; one cyst in LV and other cyst in pericardium (uncommon part) without compressive effect that appears to be remnant

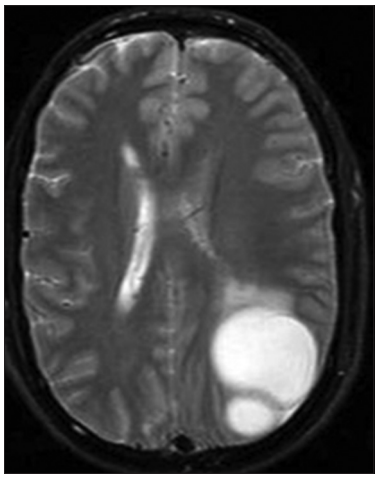

Figure 1: Brain hydatid cyst in left parietotemporal lobes (magnetic resonance imaging, axial cut)

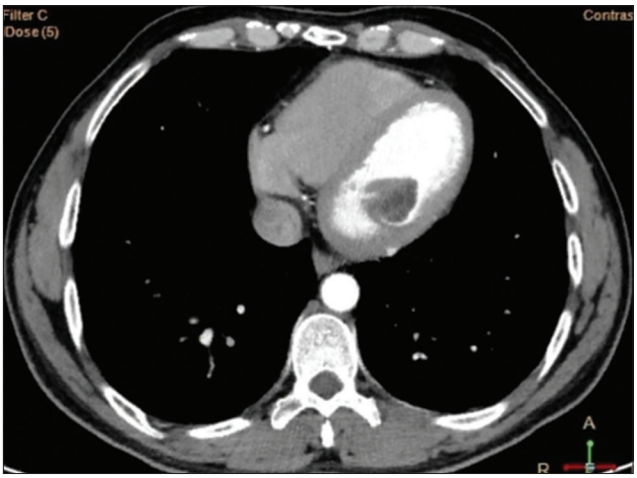

Figure 2: Cardiac hydatid cyst (computed tomography scan with contrast, axial cut)

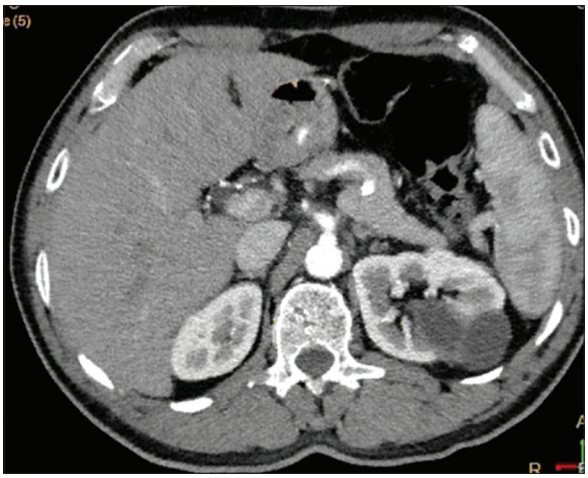

Figure 3: Left renal hydatid cyst (computed tomography scan with contrast, axial cut)

of the previous operated cyst. In cerebral HC, involvement of supratentorial in parietal lobe is common and in this case, a cyst was found in this site. Most of the renal HC cases like our case are asymptomatic and diagnosed by imaging. An important differential diagnosis in asymptomatic renal HC is poly-cystic kidney. In addition, the abscesses and tumors are the considerable differential diagnosis for HC and should be distinguished by history and para-clinic data.[5-11]

Imaging methods usually are helpful for diagnosis. Ultrasonography, computed tomography, and MRI can image the association of the cyst with neighboring tissues, the internal laminar wall of the cyst and intraluminal daughter cysts. Serologic tests can confirm that the cyst is hydatid in origin and are also useful for follow-up, although it should be noted that such tests can be false positive in up to 33% of cases.[12,13]

As soon as HC is diagnosed, treatment should be initiated promptly to prevent the serious complications. The two main treatment modalities include surgical and pharmaceutical methods. Depending on the patient's clinical condition and the characteristics of the cyst, such as its size and location, the treatment modality will be selected. Surgery is the choice treatment for brain, kidney and cardiac HCs, but there may be anaphylactic shock and spread to multiple organs, which can pose a great challenge in its eradication.[14,15] Unfortunately, our case refused surgical treatment because of these complications and followed medical treatment.

Routinely when radiologists find multiple cystic lesions at different organs or a cystic lesion suspected of HC in an atypical site, especially in an endemic area, they usually examine liver and lung as the most common involved organs in HC. If liver and lungs were not infected, radiologists likely rejected HC diagnosis; while in our case, rare sites involvement was seen and common organs were not infected.

The limitations of this plan include the patient's lack of cooperation for further diagnostic procedures and the lack of referral for follow-up. Also, the patient refrains from performing surgery in the last episode of recurrence.

CONCLUSION

Our case is an example of precision in the diagnosis of HC; a case from an endemic area with multiple cystic lesions in three uncommon sites for HC (brain, heart and kidney) and in different periods of time; while liver and lungs as common HC sites were intact. In this situation, HC should not be ruled out in reporting of the imagines and more investigations like more specific and sensitive serologic tests and correlation with history, clinical findings and epidemiology for HC should be performed. Because accurate and timely diagnosis of HC and a proper approach include appropriate medical or surgical procedures can reduce HC morbidity and mortality.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Acknowledgement

We would like to thank the Clinical Informationist Research Center dependent on Deputy of Research and Technology of Isfahan University of Medical Sciences

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Colledge NR, Walker BR, Ralston SH. Davidson's Principles and Practice of Medicine. Edinburgh, Scotland: Churchill Livingstone Elsevier; 2010. p. 329.
2. Mandal S, Mandal MD. Human cystic echinococcosis: Epidemiologic, zoonotic, clinical, diagnostic and therapeutic aspects. Asian Pac J Trop Med 2012;5:253-60.
3. Prousalidis J, Tzardinoglou K, Sgouradis L, Katsohis C, Aletras H. Uncommon sites of hydatid disease. World J Surg 1998;22:17-22.
4. Sachar S, Goyal S, Goyal S, Sangwan S. Uncommon locations and presentations of hydatid cyst. Ann Med Health Sci Res 2014;4:447-52.
5. Aleksic-Shihabi A, Vidolin EP. Cystic echinococcosis of the heart and brain: A case report. Acta Med Okayama 2008;62:341-4.
6. Gupta M, Singh R, Lehl SS. Isolated renal hydatid cyst in a diabetic postmenopausal female. J Midlife Health 2015;6:134-6.
7. Kızılca O, Altaş M, Senol U, Oztek MA. Hydatid disease located in the cerebellomedullary cistern. Case Rep Med 2014;2014:271365.
8. Erşahin Y, Mutluer S, Güzelbağ E. Intracranial hydatid cysts in children. Neurosurgery 1993;33:219-24.
9. Ozdemir NG, Kurt A, Binici DN, Ozsoy KM. *Echinococcus alveolaris*: Presenting as a cerebral metastasis. Turk Neurosurg 2012;22:448-51.
10. Capoğlu I, Unüvar N, Erdogan F, Yilmaz O, Caydere M. A hydatid cyst of the thyroid gland. J Int Med Res 2002;30:206-9.
11. Rahman A, Yücel A, Yilmaz M. A case of secondary localized pericardial cyst hydatic disease and determination of cyst hydatic scoleces and hooks with various stains. Turkiye Parazitol Derg 2008;32:31-4.
12. Brunetti E, Kern P, Vuitton DA, Writing Panel for the WHO-IWGE. Expert consensus for the diagnosis and treatment of cystic and alveolar echinococcosis in humans. Acta Trop 2010;114:1-6.
13. Biava MF, Dao A, Fortier B. Laboratory diagnosis of cystic hydatic disease. World J Surg 2001;25:10-4.
14. Shehatha J, Alward M, Saxena P, Konstantinov IE. Surgical management of cardiac hydatidosis. Tex Heart Inst J 2009;36:72-3.
15. Shahait M, Saoud R, El Hajj A. Laparoscopic treatment of giant renal cystic echinococcosis. Int J Infect Dis 2016;42:58-60.