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Case Report

Isolatedextra Hepatic and Extra Pulmonary Hydatid Cyst: Report of 33 Rare Cases

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Abstract

Background: Hydatid disease is a parasitic infestation caused by *Echinococcus granulosus* and caused by the larval stage of the Echinococcus tapeworm and it primarily affects the liver and lung but involvement of other organs is also possible secondary to peritoneal seeding or hematogeneous dissemination. About extra hepatic and pulmonary hydatid cyst (EHPHC) there are not basic studies. we want to discuss this entity and review the literatures.

Materials and methods: A systematic retrospective review was performed for all extra hepatic and pulmonary hydatidcyst (EHPHC) patients treated in our province between 1990and 2016. Inclusion criteria in this study, was all patients with (EHPHC) organ involvement without simultaneously involvement liver and lung by hydatid cyst. Final diagnosis confirmed with pathologist. The patient's records were studied for gender, age, site of involvement, diagnosis and results of treatment. All data was analyzed by using SPSS version 21.

Results: In this study record of 33 patients who were evaluated within this period, were reviewed. 21 patients were male and 12 patients were female. Age of patients was 25 to 56 years old. Hydatid cyst of spleen was the most common all of (EHPHC). Others were Kidney, pancreas, soft tissue and mediastinum. The most common tools for diagnosis was U/S and CT-scan. All patients with hydatid cyst of spleen underwent splenectomy. Others (EHPHC) underwent radical resection. Albendazole used in all patients just post operation. Outcomes of surgery were good and no recurrences were occurred.

Conclusion: Although liver and lung are the most commonly involved organs, in 10% of cases it occurs in other locations. Hydatid cysts should be included in the differential diagnosis of any cystic mass of all organs in body, especially in endemic regions. Total remove of the cyst component without any spillage is the best treatment option.

INTRODUCTION

Hydatid cyst (HC) is a zoonotic infection caused by *Echinococcus granulosus*, or less frequently, *E. multilocularis* which occurs worldwide [1,2]. However, (HC) remains a considerable public health problem in several mediterranean countries [3,4]. The dog or other carnivore is the definitive host while sheep is the intermediate host [3,4]. The adult worm lives in the intestine of the definitive host and discharges eggs in their feces [1,4]. The intermediate host ingests the ovum while vegetable on contaminated ground. The developed embryo, passes through the intestinal wall to enter the portal venous system and embedded in the liver which is the first organ most frequently involved [4,5]. The life cycle is completed when an infected intermediate host dies and their viscera which contain the larval are consumed by a definitive host [3,4]. Man becomes infected by contact directly with a definitive host or by eating contaminated water or vegetables [1,4,5]. (HC) disease has been involve almost every organ of the human body, but approximately 70% of the cysts are involve in the liver, followed by the lung (15%–47%) and 10% rest of body [2-4]. The disease is less frequently found in the spleen, pancreas, heart, brain, kidney, bones, adrenal and muscles, soft tissue and rib [1,2,4,5,6,9]. The liver or lung involvement may by rollout of (HC), if a cyst is present where else in the body [1-3,5]. The fluids in the cyst are crystal and clear [2,4,5]. It is transudate of serum contains protein and may be high antigenicity [5,6]. As far for diagnosis of (HC) according serology tests have little role, due to its high false positive or false negative rate up to 15% - 20% [6]. Ultrasonography and CT scan provide characteristic appearances for diagnosis especially in hepatic disease while extra hepatic disease may pose a diagnostic challenge [6]. The suspected diagnosis was confirmed by histopathological examination [5-7]. The choice of treatment is surgery for eradication and diagnosis of (HC). The aim of this study is to discuss presentation, diagnosis and treatment of extrahepatic and pulmonary hydatid cyst, because this cyst can affect any part of the body and any age groups, except the hair and nails with various symptoms and sign and is challenging [3,8-10].

METHOD AND MATERIALS

In this retrospective study, we reviewed the data of 33 patients with extra hepatic and extra pulmonary hydatid cyst (EHPHC) In Guilan province, Razi, Arya and Golsar Hospitals, Inflammatory, Lung Disease Research Center, Guilan University of Medical Sciences, Rasht, Iran between 1990 to 2016. Inclusion criteria in this study, was all patients with (EHPHC) organ involvement without simultaneously involvement liver and lung by hydatid

cyst. The data including symptoms, signs and imaging's feature and surgical approaches (Table 3), and outcome of surgery. Ultrasonography and CT scan was performed in all patients because this imaging, provide characteristic appearances for diagnosis especially in hepatic disease while extra hepatic disease may pose a diagnostic challenge [6] and MRI was used in soft tissue cysts. Serology tests were not performed routinely. Because has little role to help for diagnosis, due to its high false positive or false negative rate up to 15% - 20% [1,6]. In three of soft tissue (EHPHC) needle aspiration and biopsy was performed and was diagnostic and in others the diagnosis was obtained intra-operatively [7-9]. All patients treated with albendazole 10 mg/kg twice a day in three cycle of 28 day with 14 day interval in early postoperatively. Data were collected forced of patients and analyzed by using SPSS version 21.

Figure 1 Shows hydatid cyst of spleen.

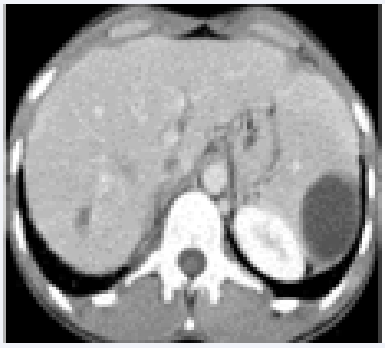

Figure 2 CT scan show cystic lesion of spleen.

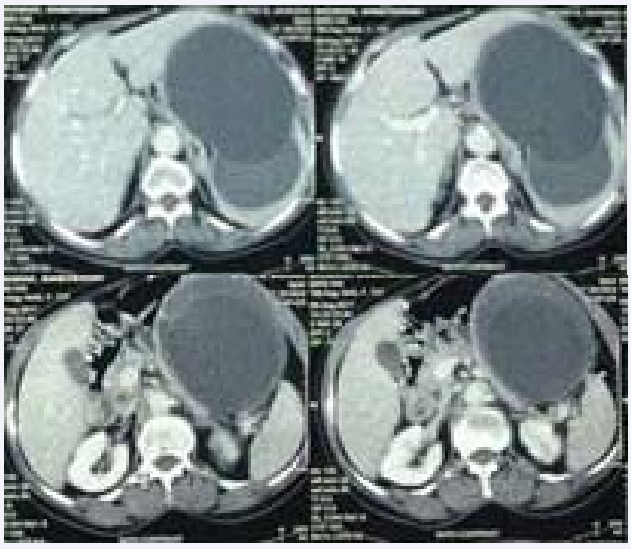

Figure 3 Show hydatid cyst of pancreas.

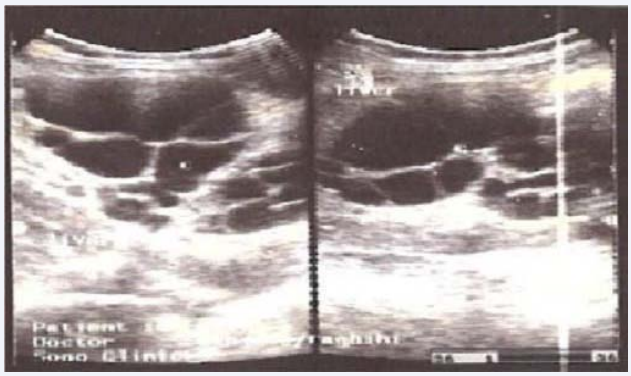

Figure 4 U&S of kidney shows hydatid cyst.

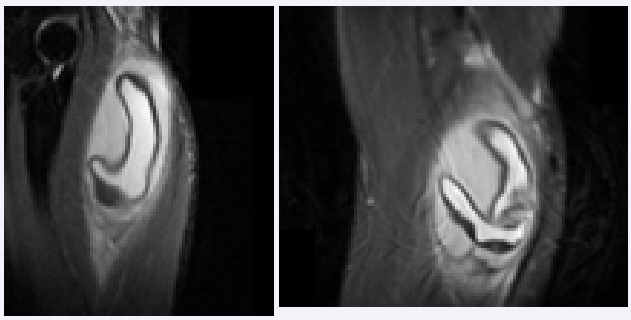

Figure 5 MRI show hydatid cyst of brachioradialis muscle.

Figure 6 U&S of right axilla show hydatid cyst.

RESULTS

21of patients were male and 12 of them were female. The average age at the time of the treatment was 34 years and ranged from 12 to 58 years. Eight of patients present with abdominal masses. The most common organ involvement was spleen, pancreas and kidney. The distribution of organ involvement was shown in Table 1. Four of patients present with left upper quadrant pain due to spleen hydatid cyst. Two of patients present with a long history mass with pain and difficulty in shoulder movements

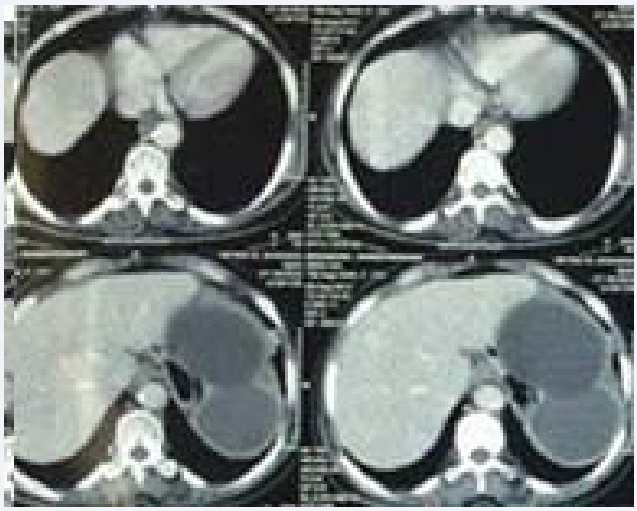

Figure 7 CT scan shows primary hydatid cyst of stomach.

Figure 8 CTscan of chest shows hydatid cyst of rib.

due to axillary region and trapezius muscle cyst, other symptoms and signs were shown in Table 2. Ultrasonography and CT scan was the most common tools for diagnostic. MRI used in two patient with brachioradialis muscle and cervical cyst (Figures 5,11). Three of patients with soft tissue cystic mass to underwent needle aspiration biopsy and was diagnostic. All patients underwent surgical resection. In four spleen (HC) splenectomy was the surgical procedure and in two cases evacuation and per cystectomy was performed. Surgical approach in two cases with pancreatic (HC) was puncture, evacuation and cystojejunostomy and in one case distal pancreatectomy was performed. In two cases with (HC) of kidney (Figure 4), the surgical method was puncture and pericystectomy and capitonnage and in one case nephrectomy was performed. In mediastinal cyst we performed cyst aspiration and evacuation and pericystectomy. Inrib (HC), aspiration, evacuation, pericystectomy and rib resection was performed. In rest organ, we performed puncture, evacuation and pericystectomy. We have not any morbidity and mortality. In average five year follow-up there was not recurrence.

DISCUSSION

Hydatid cysts (HC), primarily affects the liver and lung.The extra hepatic disease is considered to be the consequence of a secondary infection via metastatic spread from the hepatic focus [6,10,11], (HC) has been involve almost every organ of the human body, but approximately 70% of the cysts are involve in the liver, followed by the lung (15%–47%) and 10% involve rest of the body [2-4]. Spleen, pancreas, heart, brain, kidney, bones, adrenal glandmuscles, soft tissues and rib may frequently found in (HC) [1,2,4,5,9]. Isolated extra hepatic hydatid without hepatic and pulmonary involvement is a rare phenomenon, the explanation given are hepatic omission and passage of oncospheres through hepatic sinuses without carrying disease, and passage via lymphatic vessels or via portocaval anastomosis and the vascular passage in a retrograde fashion [6,11]. Splenic involvement in (HC) is uncommon, representing less than 2% of all human involved by Echinococcus [12]. In this study we revived six case of primary splenic cysts. Splenic (HC) may be primary or secondary to ruptured (HC) of liver [13]. Possible routes of primary (HC) of spleen include arterial route after passing through liver and lung [9]. The differential diagnosis includes cystic lesions of adjacent organs as Pancreas, liver and omentum, intrasplenic aneurysm [14]. Ultrasonography and CT-scan, are the most valuable imaging tools for the diagnosis and evaluation of splenic diseases as our cases (Figures 1,2) [14]. Total splenectomy, partial splenectomy, cyst evacuation and pericystectomy with omentoplasty are the surgical techniques to treat splenic (HC) [14]. In our cases, total splenectomy, aspiration, evacuation of cyst and pericystectomy was the preferred methods for splenic (HC). Chemotherapy and Puncture, Aspiration, Injection, and Re-aspiration (PAIR) is another newer technique for splenic (HC) [12,14]. We did not used this method. Hydatid cyst of the pancreas (PHC) is rare, since it accounts for less than 1%, even in countries where (HC) is endemic [15]. In this series we treated three (PHC) (Figure 12). The clinical presentation of (PHC) are, obstructive jaundice, fistulization and spillage of scolices into the biliary tree and acute pancreatitis [16]. Cysts located in the body and tail of pancreas may present with abdominal pain, vomiting, fullness and early satiety, due to compression of the stomach by cyst [15,17]. Two cases of our patients presented with epigastric pain and in one case with (PHC) of tail and body of pancreas which presented with back pain (Figure 3). The diagnosis of (PHC) can

Figure 9 show omentumhydatidcyst.

Figure 10 CT scan show hydatid cyst of mediastinum.

be performed by ultrasonography, CT scan, MRI (Figures 5-10) and endoscopic ultrasound. The radiological imaging features are useful for distinguishing hydatid cyst from other cystic lesions of pancreas [15-17]. We used ultrasonography and CT scan in our patient. Surgery remains the treatment of choice in (PHC). Partial or total cystectomy, marsupialization and external drainage have also been reported in the management of pancreatic

Figure 11 MRI of neck show hydatid cyst in two views (subcutaneous).

Figure 12 show pericardial hydatid cyst.

Table 1: Organ distributions of (HC) in 33patients.
Organ No
Spleen 6
pancreas 3
kidney3
omentum 2
rib 2
mediastinum 2
muscles(Tr,Qu,Br)* 3
breast 1
axilla 1
inguinal 1
chest wall 1
retroperitoneal1
incision site(CW)** 1
incision site(AW)*** 1
meso of intestine 1
cervical spine 1
occiput region(SC)**** 1
stomach 1
pericardium 1
*Trapezius,Quadriceps,Brachioradialis
**chest wall
***abdominal wall
****subcutaneous

Table 2: Show Symptoms and sign in 33 case with (HC).
Organ symptoms sign
Spleen Pain incidental(no=2)
Soft tissue pain mass
Kidney pain. hematuria mass
Pancreas pain mass
Rib pain mass
Mediastinum pain ---
Abdomen wall pain mass
Chest wall pain mass
cervical pain Horner SY*
occipitalpain mass

Table 3: Surgical approach in 33 case with(HD).
Spleen splenectomy(n=4) AEO*(N=2)
Pancreas AECJ** distal pancreatectomy (n=1)
KidneyPPC***(N=2) nephrectomy(n=1)
Muscles radical resection ----------
Rib evacuation ,rib resection ----------
mediastinum radical resection ----------
chest wall radical resection ----------
abdominal wall radical resection ----------
others radical resection ----------
*AEO=aspiration,evacuation,omentoplasty
**aspiration,evacuation, cystojeujonostomy
***puncture,pericystectomy,capitonnage

hydatidcysts, [15-18]. Many surgical procedures are available to remove the cyst. We used cystojejunostomy in two cases without any complication and distal pancreatectomy in one case. We do not use any scolex agent in the cavity of cyst because risk of fatal pancreatitis is high. In external drainage long time discharge may occur and we don't recommended [19,37].

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REFERENCES

1. Aghajanzadeh M, Safarpoor F, Amani H, Alavi A. One-stage procedure for lung and liver hydatid cysts. Asian Cardiovasc Thorac Ann. 2008; 16: 392-395.
2. Agayev RM, Agayev BA. Hepatic hydatid disease: Surgical experience over 15 years. Hepatogastroenterology. 2008; 55: 1373-1379.
3. Qassim F, Munther I. Surgical considerations in hydatid disease: Clinical Surgery a practical guide. 2009; 376-389.
4. Ahmed AE. Taki-Eldin. A solitary primary subcutaneous hydatid cyst in the thigh: a case report. Int Surg J. 2016; 3: 411-414.
5. Yardimci S, Ulas M, Surmelioglu A, Bostanci EB. Splenic recurrence of liver hydatid cyst and spleen preserving therapy. Singapore Med J. 2011; 52: e2232-e225.
6. Rauniyar RK, Sharma U, Baboo S. Isolated Extra hepatic Hydatid Cyst of Para spinal Muscle - Unusual Presentation - a Case Report. NJRI. 2012; 2.
7. Manoucheher Aghajanzadeh, Mehdi Karimian, Zahra Sadat

Segatoleslami, ShirinManshori, RassolHassanzadeh, TaherehMarasi. Primary Isolated Hydatid Cyst in Trapezes Muscle: A Extremely Rare Site. Ann Clin Pathol. 2017; 5: 1111.

8. Guraya SY, Alzobydi AH, Guraya SS. Primary extra hepatichydatid cyst of the soft tissue: a case report. Journal of Medical Case Reports. 2012; 6: 404.

9. Aghajanzadeh M, Alavi CE, Asgary MR, Rimaz S, Massahnia S. Primary Hydatid Cyst of the Rib: Present as a Chest Wall Mass. MOJ Surg. 2016.

10. Manouchehr Aghajanzadeh, Sina Khajeh, jahromi, RasoolHassanzadeh, HannanEbrahimi. Posterior Mediastinal Cyst. Arch Iran Med. 2014; 17: 95-96.

11. Von Sinner WN, Akhtar M. Primary spinal echinococcosis (Echinococcusgranulosus) of lumbosacral spine with destruction of the left pedicles of L3-5 and extension of a large paraspinal cystic mass into the spinal canal. Skeletal Radiol. 1994; 23: 220-223.

12. Gupta V, Kaira V, Sharma J, Sen R, Sangwaiya A. Primary Hydatid Cyst of Spleen: A Rare Entity. J Trop Dis. 2014; 2: 131.

13. Rasheed K, Zargar SA, Telwani AA. Hydatid cyst of spleen: a diagnostic challenge. N Am J Med Sci. 2013; 5: 10-20.

14. Williams RJ, Glazer G. Splenic cysts: changes in diagnosis, treatment and aetiological concepts. Ann R Coll Surg Engl. 2016; 75: 87-89.

15. Zeeshan Ahmed, SanjeevChhabra, Ashish Massey, VikeshVij, Rahul Yadav, RajendraBugalia, et al. Jenaw'Primary hydatid cyst of pancreas: Case report and review of literature. Int J Surg Case Rep. 2016; 27: 74-77.

16. Eckert J, Deplazes P. Biological, epidemiological, and clinical aspects of echinococcosis, a zoonosis of increasing concern. Clin Microbiol Rev. 2004; 1: 107-135.

17. Masoodi MI. Hydatid cyst of the pancreas: a case report and brief review. Turk J Gastroenterol. 2011; 22: 430-432.

18. Karaman B. Percutaneous treatment of a primary pancreatic hydatid cyst using a catheterization technique. Korean J Radiol. 2012;13: 232-236.

19. Venkatesh Seetharam, Vinay Khanna, Padmapriya Jaiprakash, Kranthi Kosaraju, Joseph Thomas, Chiranjay Mukhopadhayay. Case Primary Hydatid Cyst of the Kidney and Ureter with Hydatiduria in a Laboratory Worker: A Case Reports in Nephrology. 2012 (2012), Article ID 596923, 3 pages.

20. Gogus C, Safak M, Baltaci S, Turkolmez K. Isolated renal hydatidosis: experience with 20 cases. J Urol. 2003; 169: 186-189.

21. Mohamed Rami, khalid Khattala, Aziz ElMadi, My Abderrahmane Afifi, Youssef Bouabddallah. The renal hydatid cyst: report on 4 cases. Pan Afr Med J. 2011; 8: 31.

22. Mohammad Sadegh Eamaeili Delshad, Manoucheher Aghajanzade, Siamakrimaz. Hossein Hemmatiand Reza Shojaee: Complicated primary Hydatid Cyst in brachioradialismuscle: An extremely unusual site: Merit Research Journal of Medicine and Medical Sciences. 2017; 4: 076-077.

23. Kumar A, Kumar A, Gaurav K. A rare case of isolated cyst of breast. Int J Surg Case Rep. 2015; 7:115-118.

24. Mirdha BR, Biswas A. Echinococcosis: presenting as palpable lumps of the breast. Indian J chest. 2001; 43: 239-241.

25. V. Abhishek, Vijayraj S. Patil, Ullikashi Mohan, BS. Shivswamy. Abdominal Wall Hydatid Cyst: Case Report and Review of Literature. Case Rep Surg. 2012; 2012: 583294.

26. Ozsoy M, Keles C, Kahy M, Keles G. Primary echinococcal cyst in the axillary region. J Infect Dev Ctries. 2011; 5: 825-827.

27. Mehrangiz Zangeneh, Mahmood Amerion S, Mohsen Alijani. Primary Hydatid Cyst of the Axillary Region: A Case Report. Case Rep Med. 2012; 2012: 36261028.

28. Suleyman Caglar Ertekin, Tolga Ozmen. Primary Hydatid Cyst of the Small Intestine: A Rare Case Report and Brief Review of the Literature. Cureus. 2016; 8: 7.

29. Sajad A Wani, Aejaz A Baba, Nisar A Bhat, Raashid Hamid, GowherNMufti. Inguinal Hydatid cyst in a child: A rare case report. Int J Surg Case Rep. 2015; 10: 236-237.

30. Orhan Z, Kara H, Tuzuner T, Sencan I, Alper M. Primary subcutaneous cyst hydatic disease in proximal thigh: an unusual localisation: a case report. BMC Musculoskelet Disord. 2003; 4: 25.

31. Ahmed AE. Taki-EldinA. Solitary primary subcutaneous hydatid cyst in the thigh: a case report. International Surgery Journal. 2016; 3: 411-414.

32. Manouchehr Aghajanzadeh, Cyrus Emir Alavi, Mohammad Reza Asgary, SiamakRimaz, Sara Massahnia. Primary Hydatid Cyst of the Rib: Present as a Chest Wall Mass. MOJ Surgery. 2016; 3.

33. Grigorios Tsigkas, Konstantinos Chouchoulis, Efstratios Apostolakis, Christina Kalogeropoulou, Nikolaos Koutsogiannis, Dimitra Koumoundourou, et al. Heart echinococcus cyst as an incidental finding: early detection might be life-saving. Journal of Cardiothoracic Surgery. 2010; 5: 124.

34. Aghajanzadeh M, Molaie R, Aghajanzadeh H, Marandi KF. Hydatid Cyst as a Cause of Pancoast's Syndrome. Arch Iran Med. 1999; 2.

35. Nur Dikmen Yaman, Mustafa Sirlak. Cardiac Hydatid Cysts- Review of Recent Literature. J Vet Med Res. 2014: 1102.

36. Ilica AT, Kocaoglu M, Zeybek N, Guven S, Adaletli I, Basgul A, et al. Extrahepatic abdominal hydatid disease caused by Echinococcusgranulosus: imaging findings. AJR Am J Roentgenol. 2007; 189: 337-343.

37. Manouchehr Aghajanzadeh. PrimaryHydatid Cyst of the Stomach: A Rare Case Report and Review of the Literature. J Liver Res Disord Ther. 2017: 3: 1-7.

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