Contents lists available at ScienceDirect

IDCases

journal homepage: www.elsevier.com/locate/idcr

Case report

A novel method for treatment of pseudomonas pyogenic hepatic abscess complicating an echinococcal cyst by irrigation with acetic acid. A case report and literature review

Kowthar Salman Hassan

Sultan Qaboos University Hospital, Department of Medicine, Oman

ARTICLE INFO	A B S T R A C T
Article history: Received 13 March 2021 Accepted 9 June 2021	 Hepatic hydatid cysts can be complicated by pyogenic abscesses. We report a case of hepatic hydatid cysts presenting to Sultan Qaboos University Hospital (SQUH) on 29 June 2010 complicated by secondary infection with fully sensitive <i>Pseudomonas aeruginosa</i>. The infection persisted despite standard treatment (drainage and systemic antibiotics) and was eventually cured with a novel method using acetic acid. <i>Methods</i>: Instillation of acetic acid (in the form of white vinegar) into the hepatic abscess through the pig tail drainage tube. <i>Results</i>: The administration of acetic acid (6%) in the form of white vinegar was well tolerated and safe to the patient and resulted in complete clinical and radiographic resolution of the hepatic abscess. <i>Conclusion</i>: Acetic acid (vinegar) irrigation can be used to clear a refractory pyogenic hepatic abscess due to<i>P aeruginosa</i> infection.
<i>Keywords:</i> Hydatid cyst Pseudomonas aeruginosa Liver abscess Acetic acid White vinegar Biofilms	

© 2021 The Author. Published by Elsevier Ltd. This is an open access article under the CC BY-NC-ND licenses (http://creativecommons.org/licenses/by-nc-nd/4.0/).

Introduction

Human hydatid disease caused by *Echinococcus granulosus* is a zoonotic infection that is highly endemic in South America, China, Africa, and Mediterranean countries [1].

Treatment of hepatic hydatid disease is with albendazole and either cystectomy or puncture aspiration, injection, re-aspiration (PAIR) procedure [2,3].

Secondary infection of the Echinococcus cyst by various bacteria and fungi is a common complication of hydatid disease and the standard treatment of such infections is with systemic antibiotics and drainage [4–7].

Here, we report a case of hepatic hydatid cysts that were treated with PAIR procedure and albendazole but complicated by superimposed pyogenic abscess due to *P aeruginosa*. This infection persisted despite appropriate antibiotics and percutaneous drainage. It was finally treated with a novel method using irrigation of the abscess with acetic acid in the form of white vinegar through the drainage tube.

Case report

A 40-year-old previously healthy lady from Bahla in Oman was referred to Sultan Qaboos University Hospital on 29 June 2010 from a private hospital with a six months history of right upper quadrant (RUQ) pain and intermittent undocumented fevers and an abdominal CT scan showing a large liver cyst. There was no history of travel, consumption of undercooked meat or raw dairy products and no animal contact. Examination revealed pain and tenderness in the RUQ and temperature of 37.9oC. Her laboratory results were unremarkable. An MRI Liver done at our hospital showed two well defined lobulated lesions in the liver measuring 13 \times 7.7. \times 10.8 and 4.7 \times 3.8 cm located in segment 4 predominantly with extension to other lobes (right cyst) and in segment 2 (left cyst) respectively. Both lesions were hypo-intense on T1 and hyper-intense on T2, had a capsule with post contrast enhancement and neither had septations. The MRI features were suggestive of hydatid cyst. See Fig. 1.

This was supported by Echinococcus serology done by ELISA test (Cellognost, Siemens) that was strongly positive at a titer of 4096.

She was started on albendazole 400 mg bid, underwent US guided PAIR procedure of the left cyst (320 mL of fluid was aspirated, hypertonic saline 20 % injected into the cyst, left for 15







E-mail addresses: kowsan@squ.edu.om, kowhassan@gmail.com (K.S. Hassan).

http://dx.doi.org/10.1016/j.idcr.2021.e01186

^{2214-2509/© 2021} The Author. Published by Elsevier Ltd. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).



Fig. 1. a and b. MRI (T2 images) of liver showing 2 cysts in segments 2 and 4.



Fig. 2. CT scan post PAIR showing 2 cysts, one with detached membranes.

min then aspirated. Finally, 100 mL of 99 % alcohol was infused into the cyst and left for 20 min before aspiration). The procedure was done in the radiology department by intervention radiologist under aseptic conditions. Cefuroxime was started post PAIR in response to a spike of fever. Aspirated fluid showed scolices of hydatid cyst further confirming the diagnosis of hydatid disease.

Post PAIR CT showed significant decrease in size of the cyst with multiple pockets of gas and membrane detachment was seen. See Fig. 2.

She was discharged on day 14 (of admission) on albendazole for 28 days and cefuroxime for 7 days and with a drain in situ with frequent follow ups. The drain was removed 6 weeks post PAIR.

A CT scan done approximately 4 month post PAIR showed marginal increase in the size of the hepatic cysts. 3.8×6.8 cm from 3.5×6.5 cm of the left cyst and 6.7×5.7 cm from 5.6×4.3 cm of the right cyst. See Fig. 3. The patient was readmitted and had PAIR of second cyst (segments 4/8). This drained thick yellow pus although samples sent to microbiology laboratory showed no growth. Pus thickness required frequent flushing of the draining tube with saline.

The patient was discharged on day 8 of admission, with the drain and on albendazole and moxifloxacin with regular follow ups. One week later the drain had 50 mL of fluid that grew



Fig. 3. CT scan showing marginal increase in both the size of cysts.

Pseudomonas aeruginosa despite her being discharged on moxifloxacin. Her Echinococcus serology here was < 16. Albendazole was, therefore, stopped and moxifloxacin was changed to ciprofloxacin 500 mg b.i.d for 3 weeks. This second drain was removed one-month post insertion after cessation of any drainage.

Six weeks post removal of drain 2, she presented with pus discharge from both previous drain sites and burning pain at the site disturbing her sleep. An urgent CT showed decrease in the size of both cysts (3.6×4.3 cm from 3.7×4.5 cm in the left cyst and 3×2.5 cm, from 6.8×5 cm in the right cyst). The left cyst had tracts connecting it to the abdominal wall. She was therefore, admitted on 29/1/2011 and treated with Tazocin 4.5 mg t.i.d plus iv ciprofloxacin 400 mg b.i.d. The residual fluid was not drainable as advised by the intervention radiologist. Abdominal US done 14 days after this admission showed small residual debris in the cavity and the patient was discharged on ciprofloxacin 750 b.i.d after 2 weeks of iv antibiotics in hospital.

A Follow up CT abdomen done at 1 and 2 month after last admission showed unchanged size of lesion in the right lobe of the liver and significant decrease in the size of the left lesion. Ciprofloxacin was stopped 2 months after last admission. Five months post last follow up, the patient attended the OPD with 7 days of RUQ pain on deep breathing, coughing or sneezing and on lying on R side. An urgent CT showed a sub-diaphragmatic collection measuring 8.6 \times 7.5 cm and an adjacent smaller collection of 3.3 \times 2.3 cm with a subcutaneous sinus tract. See figure

Ultra sound guided aspiration was done by intervention radiologist under aseptic technique and 60 mL of pus aspirated. Tazocin 4.5 mm t.i.d was started after sample collection. Pus grew *P. aeruginosa* (sensitive to ciprofloxacin and Tazocin). No scolices were seen in the fluid and the Echinococcus serology was < 16. A follow up US one week later showed reduction in the size of the collections (3.6×3 cm from 8.7×7.5 cm and 2×1.4 cm from 3.3×2.3 cm). However, the drain continued draining 100–150 ml of pus daily for 5 weeks. The draining tube required flushing with sterile saline due to the stickiness of the pus. Due to the prolonged stay in hospital, the patient started to feel physically and psychologically exhausted and was longing for discharge especially that the month of Ramadhan (a holy month of fasting) was approaching.

After thorough review of her condition, we discussed with the patient the option of using of acetic acid in the form of white vinegar to irrigate the abscess due to failure of the conventional standard treatment of the abscess with drainage and appropriate antibiotics. We clearly explained that although vinegar had been used on skin wounds, it had not been used on visceral abscesses before.

Following obtaining consent from the patient, twenty ml of white vinegar was instilled into the draining tube and withdrawn several times. This was not associated with any pain. Thick pus and debris drained out and the patient was monitored overnight for any complications. Over the next few days the drain produced debris and 10 -15 mL of serosanguinous fluid that eventually became zero. After 2 days of zero discharge (6 weeks post admission), the drain was removed and the patient was discharged on ciprofloxacin 750 b.i.d for another month.

Subsequent images done at 1 (US), 4 (CT), 8 (US), 20 (US), and 32 (US) months from last date of discharge showed normal liver with no collections. See Fig. 4. The patient remained well and asymptomatic.

Discussion

Hydatid cyst disease is a common worldwide zoonosis [1]. In Oman it is considered as one of the endemic zoonotic diseases with abattoirs being the main source of infection. A sero-epidemiological study in six municipal abattoirs revealed a 14.6 % positivity of the livestock including camels, cattle, sheep and goats [8]. Humans usually acquire the infection by consumption of contaminated soil, water or food contaminated with infected canine faecal matter.



Dogs (definite host) in turn acquire it by eating offal of infected sheep (intermediate host). Our patient was a previously healthy house wife with no contact with animals. However, consumption of food or water contaminated with Echinococcus eggs could not be entirely excluded.

Hydatid cysts can be complicated by rupture, relapse or pyogenic infections with various pathogens [4–6]. Our patient did not show any signs of rupture or hydatid relapse as her serology had dropped to < 16 and there were no scolices seen in the aspirated fluid after the initial presentation.

She, however, developed a secondary pyogenic infection caused by fully sensitive *Pseudomonas aeruginosa* that proved very difficult to eradicate. Possible sources of the secondary infection include introduction of the bacteria during PAIR procedure, during frequent flushing of the blocked tube or spontaneous. Hypertonic saline (20 %) used in PAIR does not support growth of P aeruginosa [9]. Moreover, the risk of infection of hydatid cyst post PAIR is considered to be low [10]. Frequent flushing of the draining tube afterwards with normal saline could have been the culprit as Pseudomonas is known to colonize fluids. This possibility is supported by the fact that the initial fluid at drainage of cyst 2 was negative and became positive at a later date after frequent flushing. The infection, however, could have risen de novo as reported in other cases [11,12].

Irrespective of the source of the secondary pyogenic infection, our patient received adequate conventional standard treatment for the hepatic abscess-consisting of percutaneous drainage and systemic antibiotics for sufficient duration on more than one occasion. Nevertheless, her problem persisted for 2 years.

Persistence and relapse of the bacterial infection despite the appropriate therapy (drainage and systemic antibiotics) was most probably facilitated by the formation of biofilms by the Pseudomonas which is known to form biofilms [13–15].

Biofilms comprise bacteria embedded within slimy extracellular matrix composed of polysaccharides, proteins and lipids produced by the bacteria themselves for protection [18]. Microorganisms form biofilms in response to various factors including recognition of attachment sites on surfaces, or exposure of planktonic organisms to sub-inhibitory concentrations of antibiotics [16,17]. Since our patient received adequate doses of dual antipseudomonal antibiotics (ciprofloxacin and Tazocin) for adequate duration, it is unlikely that sub-therapeutic treatment was the trigger. Instead, biofilms could have been triggered by adherence of the bacteria to residual inactive hydatid cyst.

Antibiotics that are effective against bacteria in vitro may be effective against planktonic bacteria in vivo but not so against bacterial colonies embedded within biofilms. This could well explain the failure of treatment in our case despite the use of appropriate antibiotics against *P aeruginosa*. Treatment of bacterial infections in biofilms is very challenging indeed. Electrical energy and ultrasonics have been investigated as options to eradicate biofilm infection [18,19].

We considered the use of acetic acid in this case due to the many reported medicinal uses for acetic acid including injection into tumors since 1800s, antiseptic properties when used as 1% against *Staphylococci, Streptococci, Pseudomonas, Enterococci* and in treatment of otitis externa. Its wound cleaning properties have also been noted by various cultures and for *Peduomonas* infections [20–22]. Moreover, there have been reports on its effectiveness in breaking biofilms [23,24].

White vinegar is 4–7 % acetic acid and 93–96 % water. Traditionally it was made by fermentation of foods such as sugar and potatoes but nowadays by fermentation of ethanol by addition of yeast or phosphate. It is widely used in cleaning and widely available, being cheap and easily made. Moreover, table vinegar has been used to dress wounds [25]. Vinegar used in this case was freshly opened home vinegar. Its sterility was not tested as vinegar is widely known for its antiseptic properties.

We feared induction of pain or burning sensation during vinegar instillation but were reassured by the patient denying any discomfort and reporting only sensation of cold at site of irrigation.

The effectiveness of vinegar in eradiation of the infection in this case was proved by the complete resolution of the infection clinically and radiologically over 8 years of follow up.

Conclusion

Acetic acid (vinegar) irrigation can be used to clear a refractory pyogenic hepatic abscess due to *Pseudomonas aeruginosa* infection.

We, hence, report the first case of treating *Pseudomonas aeruginosa* hepatic abscess complicating hydatid cyst with acetic acid instillation and suggest that further studies should be conducted on this method to make it more widely used in similar cases and possibly in central line lock therapies for removing biofilms.

We call the method of instillation of vinegar (acetic acid) into abscesses Hassan Procedure.

Funding

No funding is required.

Author contribution

This manuscript was written by only one author, Kowthar Salman Hassan.

Declaration

The patient and her next of kin gave consent at time of procedure. Consent for publication of the case was obtained from the patient over the phone at a much later date.

Declaration of Competing Interest

The authors report no declarations of interest.

References

- Eckert J, Conraths FJ, Tackmann K. Echinococcosis: an emerging or re-emerging zoonosis? Int J Parasitol 2000;2000(30):1283–94, doi:http://dx.doi.org/ 10.1016/s0020-7519(00)00130-2.
- [2] Alfieri S, Doglietto GB, Pacelli F, Costamagna G, Carriero C, Mutignani M, et al. Radical surgery for liver hydatid disease: a study ofconsecutive patients. Hepatogastroenterology 1997;44(14):496–500.
- [3] World Health Organization. Puncture, aspiration, injection, re- aspiration: an option for the treatment of cystic echinococcosis. Geneva, Switzerland: World Health Organization; 2001. http://apps.who.int/iris/handle/10665/67207.
- [4] Koçer NE, Kibar Y, Guldur ME, Deniz H, Bakir K. A retrospective study on the coexistence of hydatid cyst and aspergillosis. Int J Infect Dis 2008;12(3):248– 51, doi:http://dx.doi.org/10.1016/j.ijid.2007.08.005.
- [5] Hakyemez IN, Sit M, Aktas G, Tas T, Mengeloglu FZ, Kucukbayrak A. A Case of Giant Hepatic Hydatid Cyst Infected with Morganella morganii and the

Literature Review. Case Rep Gastrointest Med 2012591561, doi:http://dx.doi.org/10.1155/2012/591561.

- [6] García MB, Lledías JP, Pérez IG, Tirado VV, Pardo LF, Bellvís LM, et al. Short Report: primary super-infection of hydatid cyst-clinical setting and microbiology in 37 Cases. Am J Trop Med Hyg 2010;82(3):376–8, doi:http://dx. doi.org/10.4269/1jtmh.2010.09-0375.
- [7] Zerem E, Hadzic A. Sonographically guided percutaneous catheter drainage versus needle aspiration in the management of pyogenic liver abscess. AJR Am J Roentgenol 2007;189(3)W138–142, doi:http://dx.doi.org/10.2214/ AJR.07.2173 [PubMed] [Google].
- [8] Al Awaidy S, Al Hashami H. Zoontic Diseases in Oman: Successes, challenges and future directions. Vector Borne Zoonotic Dis 2020;20(January (1))1–9, doi: http://dx.doi.org/10.1089/vbz.2019.2458 [PubMed] [Google].
- [9] Michon A-L, Jumas-Bilak E, Chiron RI, Lamy B, Marchandin H. Advances toward the elucidation of hypertonic saline effects on Pseudomonas aeruginosa from cystic fibrosis patients. PLoS 2014;9(2)e90164, doi:http://dx.doi.org/10.1371/ journal.pone.0090174 [PubMed] [Google].
- [10] Smego Jr RA, Bhatti S, Khlaiq AA, Beg MA. Percutaneous Aspiration-Injection-Reaspiration Drainage Plus Albendazole or Mebendazole for Hepatic Cystic Echinococcosis: A Meta-analysis. Clin Infect Dis 2003;37(8)1073–83, doi: http://dx.doi.org/10.1086/378275 [PubMed] [Google].
- [11] Karavias D, Panagopoulos C, Vagianos C, Vagenas C, Rathosis S, Androulakis J. Infected Echinococcal Cyst. A Common Cause of Pyogenic Hepatic Abscess. Ups J Med Sci 1998;93(3)289–96, doi:http://dx.doi.org/10.3109/ 03009738809178554 [PubMed] [Google].
- [12] Sah R, Calatri M, Neupane S, Pudyal S, Toledo R, Acosta L. A case of Echinococcus granulosus hepatic hydatid cyst together with pyogenic liver abscess in a Nepali patient. J Parasit Dis 2020;44(June (2))472–5, doi:http://dx. doi.org/10.1007/s12639-020-01212-9 [PubMed] [Google].
- [13] Tolker-Nielsen T. Pseudomonas aeruginosa biofilm infections: from molecular biofilm biology to new treatment possibilities. APMIS Suppl 2014;(December (138))1–51, doi:http://dx.doi.org/10.1111/apm.12335 [PubMed] [Google].
- [14] Trøstrup H, Lerche CJ, Christophersen L, Jensen PØ, Høiby N, Moser C. Immune modulating topical S100A8/A9 inhibits growth of Pseudomonas aeruginosa and mitigates biofilm infection in chronic wounds. Int J Mol Sci 2017;18(June (17))1359, doi:http://dx.doi.org/10.3390/ijms18071359 [PubMed] [Google].
- [15] Hall-Stoodley L, Costerton JW, Stoodley P. Bacterial biofilms: from the natural environment to infectious diseases. Nat Rev Microbiol 2004;2(February (2)) 95–108, doi:http://dx.doi.org/10.1038/nrmicro821 [PubMed] [Google].
- [16] Tappe D, Stich A, Frosch M. Emergence of polycystic neotropical echinococcosis. Emerg Infect Dis 2008;14(February (2))292–7, doi:http://dx. doi.org/10.3201/eid1402.070742 Turk J Med Sci 2003;33:369-374.
- [17] Canda MS, Güray M, Canda LT, Astarcioglu H. The pathology of echinococcosis and the current echinococcosis problem in Western Turkey (A report pf pathologic features in 80 cases) [PubMed] [Google]. 2003.
- [18] Wook K, Sowmya Subramanian S, Gerasopoulos K, Ben-Yoav H, et al. Effect of electrical energy on the efficacy of biofilm treatment using the bioelectric effect. NPJ Biofilms Microbiomes 2015;23(September 1)15016, doi:http://dx. doi.org/10.1038/npjbiofilms.2015.16.eCollection 2015. [PubMed] [Google].
- [19] Qian Z, Sagers RD, Pitt WG. The effect of ultrasonic frequency upon enhanced killing of P. aeruginosa biofilms. Ann Biomed Eng 1997;25(January-February (1))69–76, doi:http://dx.doi.org/10.1007/BF02738539 [PubMed] [Google].
- [20] Nagoba BS, Selkar SP, Wadher BJ, Gandhi RC. Acetic acid treatment of pseudomonal wound infections – a review. J Infect Public Health 2013;6 (December 6)410–5, doi:http://dx.doi.org/10.1016/j.jiph.2013.05.005 [PubMed] [Google].
- [21] Sloss JM, Cumberland N, Milner SM. Acetic acid used for the elimination of Pseudomonas aeruginosa from burn and soft tissue wounds. J R Army Med Corps 1993;139(2)49–51, doi:http://dx.doi.org/10.1136/jramc-139-02-04 [PubMed] [Google].
- [22] Agrawal KS, Sarda AV, Shrotriya R, Bachhav M, Puri V, Nataraj G. Acetic acid dressings: finding the Holy Grail for infected wound management. Indian J Plast Surg 2017;50(3)273–80, doi:http://dx.doi.org/10.4103/ijps.IJPS-245-16 [PubMed] [Google].
- [23] Kundukad B, Schussman M, Yang K, Seviour T, Yang L, Rice SA, et al. Mechanistic action of weak acid drugs on biofilms. Sci Rep 2017;7(July (1)) 4783, doi:http://dx.doi.org/10.1038/s41598-017-05178-3 [PubMed] [Google].
- [24] Bjarnsholt T, Alhede M, Jensen PØ, Nielsen AK, Johansen HK, Homøe P, Adv Wound Care (New Rochelle) 2015;4(July (7))363–72, doi:http://dx.doi.org/ 10.1089/woind.2014.0554 [PubMed] [Google].
- [25] Morsi AE, Mustafa FM, El Tokhy A. Vinegar simple method in dressing of Pseudomonas Infected wound. Int Inv J Med Sci. 2016;3(8):143-6.