Irish Medical Journal
Established 1867

Issue: Ir Med J; Vol 113; No. 4; P61

Hepatic Cystic Echinococcosis (Hydatid Cyst) in a Six Year Old

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Abstract

Presentation
To describe a case of cystic echinococcosis (CE) in a previously healthy child and review epidemiology of CE in Ireland.

Diagnosis
A previously healthy 6 year old girl was found to have a cystic lesion in the right lobe of her liver. Serology for *Echinococcus granulosus* was positive, and radiological features were suggestive of CE.

Treatment
The patient was pre-treated with anti-helminthic medications before undergoing a liver segmentectomy to remove the cyst, and received further treatment with albendazole after surgery. Histological findings were consistent with CE due to *E. granulosus*, likely acquired during travel to continental Europe.

Conclusion
CE should be considered in the differential of children with asymptomatic cysts in the liver and/or lung, and a travel history elucidated in such cases.

Introduction

Cystic echinococcosis (CE, also known as hydatid disease) is an important zoonosis[1]. It is caused by the larval stage of the canine tapeworm *Echinococcus*, in particular *E. granulosus*. It represents a normal part of the tapeworm lifecycle, occurring typically in herbivorous intermediate hosts such as sheep and cattle. It affects humans when patients ingest tapeworm eggs after handling infected dogs or in contaminated food or water. Cysts occur most commonly in the liver or lungs. Complications can arise such as bacterial super-infection of cysts or anaphylaxis in response to echinococcal antigen after cyst rupture[2]. We describe the first Paediatric case of CE reported in Ireland and review the associated medical literature.

Case Report

A previously healthy six year old Irish girl underwent an abdominal ultrasound scan after a urinary tract infection. She was found incidentally to have a cystic lesion (57mm x 40mm x 31mm) in the right lobe of her liver. She was asymptomatic. A travel history revealed trips to Portugal, Spain, and France over the previous 5 years, and contact with animals at a petting zoo. A faecal sample collected from the family dog showed no tapeworm eggs. Her father was a cattle farmer, but she herself had limited contact with the livestock. Her physical examination was normal. Serology for *E. granulosus* was positive by ELISA with an OD of 0.625 (cut-off value 0.25) on two occasions. A CT scan

demonstrated a low attenuation lesion in the right lobe of the liver with at least one internal septation. An MRI scan demonstrated features of a late stage (WHO type CE3A - CE4) echinococcal cyst (see Figure 1)[3]. After multidisciplinary input, a decision was made to proceed with a liver segmentectomy. She was commenced on albendazole (15mg/kg/d divided q12h). Praziquantal (50-100mg/kg/d divided q8h) was added for two weeks before and after the surgical procedure. The segmentectomy was completed with no adverse effects, and CE due to *E. granulosus* was confirmed on histology. The cyst was non-viable. Albendazole was continued for three months post-surgery to consolidate treatment. The patient has remained well since, with no evidence of recurrence.

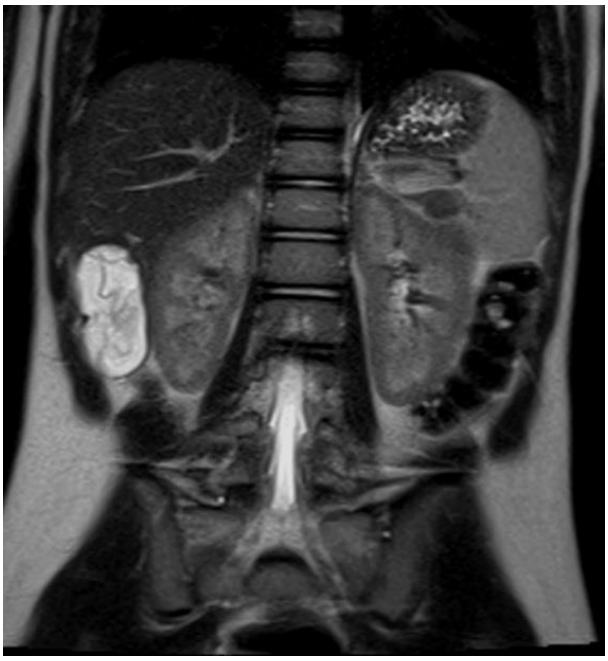

Figure 1: The T2 weighted MRI image of the liver in the coronal plane shows a single unilocular fluid containing cyst (36mm x 27mm x 51mm) in segment 6 of the liver, with an inner floating detached membrane, the so-called "water lily sign". No daughter cysts were identified.

Discussion

CE is an important zoonosis, endemic in many areas, including mainland Europe and parts of the UK. *E granulosus* strains associated with CE have not been reported in Ireland. A related strain (*E. granulosus equinus*) is endemic in Irish horses but is not known to be pathogenic to humans [4,5]. CE has been reported in Irish patients previously, typically with a history of travel to endemic region [6-8]. Ours is the first reported case of CE in an Irish child, likely acquired from travel to mainland Europe[1].

Management includes a combination of anti-helminthic chemotherapy and surgery. Treatment is determined by WHO stage, location, and number of cysts, the clinical condition of the patient, and the experience of the surgical team [9]. Surgical excision is the treatment of choice in large cysts (>10cm), cysts with daughter vesicles, and where percutaneous techniques are not available. Percutaneous techniques involve drainage of the cyst fluid and injection of scolicidal agents into the cyst cavity. PAIR (percutaneous aspiration, injection, re-aspiration) is less invasive than surgery, and can also be considered for treatment of WHO stage CE1 and CE3a cysts. Liver segmentectomy was undertaken in this case because of the location of an isolated cyst in the right lobe and the experience of our surgical colleagues. We reasoned that surgical excision would also rule out other differentials (e.g. abscess, malignancy). Leakage of cyst fluid into the abdominal cavity from either surgery or PAIR can lead to anaphylaxis [9]. Adjunctive use of anti-helminthic therapy (albendazole +/- praziquantal) reduces secondary echinococcosis. Long term follow-up (five years minimum) with serial ultrasound scans and echinococcal serology is recommended to screen for recurrence, although antibody titres suggestive of relapse/recurrence can occur even when the cyst has been successfully removed. Recurrence rates of 4%-22% have been reported [10].

In conclusion, Paediatricians should consider CE in the differential of cystic liver and lung lesions, and take a careful travel history, even in children without typical risk factors.

Declaration of Conflicts of Interest:
The authors have no conflict of interests to declare.

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References:

1. Dakkak A. Echinococcosis/hydatidosis: a severe threat in Mediterranean countries. Vet Parasitol. 2010 Nov 24;174(1-2):2-11.
2. Tinsley B, Abbara A, Kadaba R, Sheth H, Sandhu G. Spontaneous intraperitoneal rupture of a hepatic hydatid cyst with subsequent anaphylaxis: a case report. Case Reports Hepatol. 2013;2013:320418.
3. International classification of ultrasound images in cystic echinococcosis for application in clinical and field epidemiological settings. Acta Trop. 2003 Feb;85(2):253-61.
4. Hatch C. Echinococcus granulosus equinus in Irish dogs. Vet Rec. 1970 May 23;86(21):632-3.
5. Alvarez Rojas CA, Romig T, Lightowlers MW. Echinococcus granulosus sensu lato genotypes infecting humans--review of current knowledge. Int J Parasitol. 2014 Jan;44(1):9-18.
6. Butler MW, Mullan RH, Schaffer KE, Crotty TB, Luke DA, Donnelly SC. Pulmonary cystic hydatid disease in Ireland. Ir J Med Sci. 2003 Oct-Dec;172(4):204-5.
7. McFall B, Yousaf M, Calvert H, Diamond T, Epanomeritakis M. Surgical treatment of hepatic hydatid cyst. Int J Clin Pract. 2004 May;58(5):479-82.
8. O'Rourke FJ. Hydatid disease in Ireland. J Ir Med Assoc. 1969 Mar;62(381):91-3.
9. Brunetti E, Kern P, Vuitton DA. Expert consensus for the diagnosis and treatment of cystic and alveolar echinococcosis in humans. Acta Trop. 2010 Apr;114(1):1-16.
10. Prousalidis J, Kosmidis C, Anthimidis G, Kapoutzis K, Karamanlis E, Fachantidis E. Postoperative recurrence of cystic hydatidosis. Can J Surg. 2012 Feb;55(1):15-20.