

Cystic Echinococcosis: A Disease Mimicking Cancer in a Non-endemic Country

Report of Two Cases

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ABSTRACT

Echinococcosis is a parasitic disease that involves dogs as definitive host and sheep as intermediate host. Humans become infected incidentally through fecal-oral contact, particularly in the course of playful and close contact with an infected dog. Mexico is considered a region that is virtually free of cystic echinococcosis. This manuscript describes two cases that were referred to a tertiary-care oncology hospital with a diagnosis of cancer. In one case, the presumptive diagnosis was liver cancer because abdominal ultrasonography revealed a low-density mass in the right hepatic lobe. Drainage was performed and cytologic examination of the fluid showed multiple Echinococcus cyst as well as protoscolex. The case was resolved with percutaneous drainage and administration of albendazole for two months. In the second case, the patient was referred with a diagnosis of disseminated cervical cancer. A cyst was identified in the upper right lung lobe; a diagnostic puncture was performed showing an Echinococcus cyst. This resolved solely with two months of albendazole administration.

Keywords: Albendazole, cancer, echinococcosis, hydatid cyst

Equinococosis Quística: Una Enfermedad con Apariencia de Cáncer en un País no Endémico: Reporte de Dos Casos

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RESUMEN

La equinococosis, conocida también como hidatidosis, es una enfermedad parasitaria que comprende a los perros como hospedador definitivo y a las ovejas como huésped intermedio. Los seres humanos se infectan casualmente por contacto feco-oral, particularmente al jugar y estar en estrecho contacto con un perro infectado. México es considerado una región que está prácticamente libre de equinococosis quística. Este trabajo describe dos casos que fueron referidos con un diagnóstico de cáncer a un hospital oncológico de atención terciaria. En un caso, el diagnóstico presuntivo fue cáncer del hígado, porque la ultrasonografía abdominal reveló una masa de baja densidad en el lóbulo hepático derecho. Se hizo un drenaje y el examen citológico del fluido mostró quistes múltiples de equinococos así como la presencia de protoescólex. El caso se resolvió con drenaje percutáneo y administración de albendazol durante dos meses. En el segundo caso, la paciente fue remitida con un diagnóstico de cáncer cervical diseminado. Se identificó un quiste en el lóbulo superior de pulmón derecho. La punción diagnóstica realizada reveló un quiste equinocócico, que se resolvió simplemente con dos meses de administración de albendazol.

Palabras claves: Albendazol, cáncer, equinococosis, quiste hidatídico

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INTRODUCTION

Echinococcosis is a ubiquitous endemic disease, with varying prevalence and distribution influenced by agricultural, educational, economic, medical and cultural factors (1).

The *Echinococcus* life cycle involves dogs as definitive hosts and sheep as intermediate hosts (2). Certain human activities (eg the widespread rural practice of feeding dogs with the viscera of home-butchered sheep) facilitate transmission for the sheep strain and consequently raise the risk that humans will become infected. Human infection occurs through the fecal-oral route (1, 2).

The majority of primary infections in humans consist of a single cyst; however, 20–40% of individuals have multiple cysts and/or multiple organ involvement. The liver is the most common site involved, followed by lung involvement (1, 2).

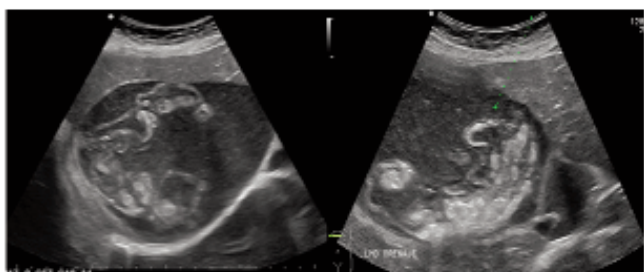
We present two patients whose referral diagnosis was liver teratoma in the first case and disseminated cervical cancer in the second.

CASE REPORTS

Case 1: A 40-year old Mexican female with abdominal pain and weight loss was referred with a probable teratoma in the liver. She lived in a suburban town in the State of Mexico and had dogs that were fed with raw viscera. One year before this, the patient was exposed to sewage water after a flood involving a river of manure in her town.

She presented six months previously with right abdominal pain, nausea, constipation and weight loss (25 kg). The only sign found on physical examination was a soft, firm and tender mass in the right inferior quadrant.

Abdominal ultrasound and computed tomography (CT) scan showed a hepatic cyst (measuring 13 × 8 × 11 cm with an estimated volume of 575 cc). A rounded hydatid lesion with a global echogenic appearance and detached membranes (water-lily sign) was observed (Figs. 1a and 1b). Drainage of



Figs. 1a and 1b: A rounded hydatid lesion with a global echogenic appearance and detached membranes (water-lily sign).

the cyst was performed; the cytopathologic study of the liquid reported the presence of multiple *Echinococcus* cysts, as well as protoscolex in the germinal layer (Fig. 2).

The patient received albendazole 400 mg twice daily for four weeks. A control ultrasound showed the cyst of the same size. A percutaneous drainage and aspiration of the

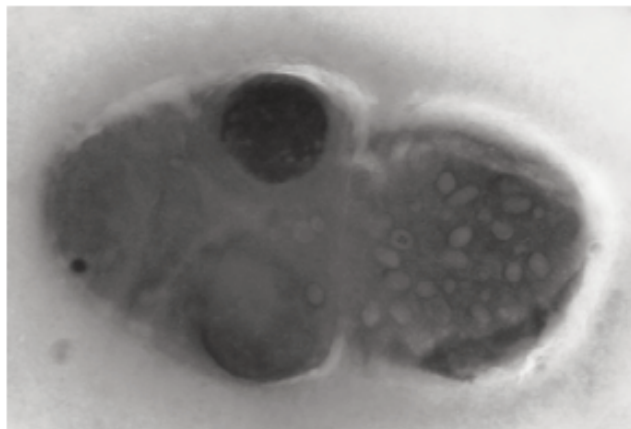


Fig. 2: Multiple *Echinococcus* cysts as well as protoscolex in the germinal layer.

liquid contents (500 cc) was performed and hypertonic saline solution was injected; a second consecutive aspiration of scolicalidal solutions was performed. The patient continued on albendazole for another month. Three months later, the patient was asymptomatic and ultrasound showed complete resolution of the lesion.

Case 2: A 66-year old Mexican female was referred with invasive cervical cancer. She lived in a rural town with limited sanitation. She had contact with a dog living outside her house that was fed exclusively with dog food.

Cervical cancer IIIB was diagnosed. She denied respiratory or systemic symptoms. Chest X-ray was performed and a 5 cm lesion located in the middle right lobe was found. Computed tomography scan indicated a 4 × 4.5 cm cyst lesion (Fig. 3). The fluid was removed by percutaneous



Fig. 3: Computed tomography (CT) scan shows a 4 × 4.5 cm cyst lesion in the middle right-lung lobe.

drainage, and the patient had a mild allergic reaction. Cytopathologic study reported multiple protoscolex compatible with *Echinococcus granulosus* (Fig. 4). The patient received albendazole 400 mg twice daily for two months. In addition to this treatment, she began radiotherapy and chemotherapy.

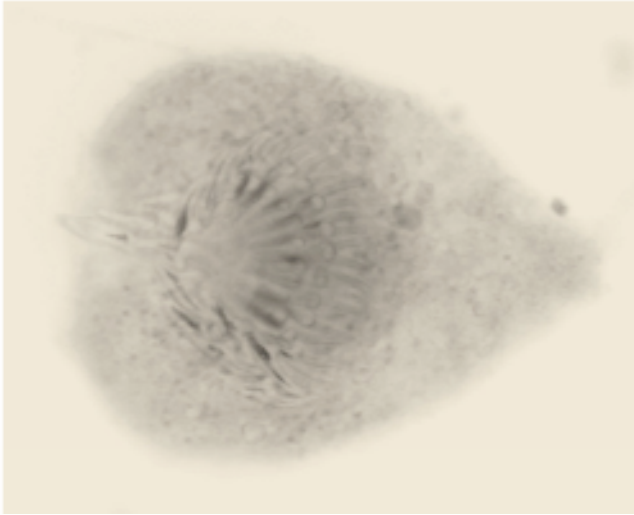


Fig. 4: Presence of multiple protoscolex compatible with *Echinococcus granulosus*.

A CT scan performed six months later showed no cyst. Two years later, the patient was asymptomatic.

DISCUSSION

Echinococcosis in humans occurs as a result of infection by larval stages of *Taenia* cestodes of the genus *Echinococcus* (3). The disease is endemic in sheep-raising areas of the world including Africa, the Mediterranean region, the Middle East, Asia, South America, Australia and New Zealand (2, 4–7). In the United States of America (USA), the majority of infections are diagnosed in migrants from countries in which echinococcosis disease is highly endemic (2, 8, 9).

In Mexico, isolation of the parasite has been accomplished in several states (10–12). There is limited information on the frequency of the disease, as only scarce data are available: 0.27–3.1% of pigs and sheep in slaughter-houses in central Mexico were positive on inspection of the infected viscera of their meat. In a suburban population residing near a slaughterhouse that detected pigs with echinococcosis, the presence of antibodies was 15%, but between 9 and 15% in test cross-reactivity with *Taenia* species (10, 13). In a community-based study performed in a rural area, prevalence was 0.75%, confirming clinical data that infection is not common (13). Humans become infected incidentally through fecal-oral contact, particularly in the course of playful and intimate contact with an infected dog (2).

Small cysts not inducing major disease may remain asymptomatic for many years, if not permanently (6). The exact time required for the development of protoscolex within the cyst in the human host is not known, but is thought to be > 10 months after infection (14). The clinical course is variable, signs and symptoms are determined by size, and cyst growth ranges from 1–5 cm in diameter per year. If a cyst ruptures, the sudden release of its contents can precipitate allergic reactions ranging from mild to fatal ana-

phylaxis (2, 7, 14). When the liver is involved, abdominal pain, nausea and abdominal mass are found (11). The main complications of hydatid cysts comprise rupture into the biliary tract, rupture into the peritoneum, rupture into the bronchial tree, and disseminated encysted echinococcosis (6, 14, 15).

Non-invasive confirmation of the diagnosis can usually be accomplished with the use of imaging and immunodiagnostic techniques. Computed tomography scan, magnetic resonance imaging (MRI) and ultrasonography are useful for the diagnosis of deep-seated lesions in all organs and also for determination of the extent and condition of avascular fluid-filled cysts (2, 3, 16).

Ultrasound findings are variable and range from purely cystic to solid-appearing pseudotumours. Wavy bands of delaminated endocyst (the water-lily sign) may be noted internally. Daughter cysts, sometimes surrounded by echogenic debris (matrix), are frequently observed. Calcifications, varying from tiny to massive, are often present peripherally. On CT, a hydatid cyst usually appears as a well-defined, hypoattenuating lesion with a distinguishable wall. Coarse wall calcifications are present in 50% of cases, and daughter cysts are identified in approximately 75% (17).

Although current gold-standard serology for human cystic echinococcosis is based on the detection of IgG antibodies in hydatid cyst fluid-derived native or recombinant antigen B subunits, sensitivity is > 95% (14, 16). Up to 10–20% of patients with hepatic cysts and about 40% with pulmonary cyst do not produce detectable specific serum antibodies, therefore giving false-negative results (14). Eosinophilia is not a frequent finding (4–15%) and is present in < 25% of infected patients (2).

Three therapeutic modalities exist to treat hepatic cystic echinococcosis: surgery, chemotherapy and percutaneous drainage (4, 18). Monitoring involves evaluation of the cyst size at three-month intervals with imaging studies and should be continued for at least three years. Changes in titre of serologic antibody values are not useful (2).

CONCLUSIONS

In Mexico, hydatosis is reported by the World Health Organization (WHO) as a “sporadic disease”, but available information suggests that this is an underestimated infection. Poor sanitation in some areas, as in the case in these women exposed to flooding sewage, places people at risk, and patients who arrive with a liver or lung mass should be included in this at-risk group. Echinococcosis must be included as a differential diagnosis to cancer.

REFERENCES

1. Filippou D, Tselepis D, Filippou G, Papadopoulos V. Advances in liver echinococcosis: diagnosis and treatment. *Clin Gastroenterol Hepatol* 2007; **5**: 152–9.
2. Moro P, Schantz PM. Echinococcosis: a review. *Int J Infect Dis* 2009; **13**: 125–33.

3. Moro PL, Schantz PM. Echinococcosis: historical landmarks and progress in research and control. *Ann Trop Med Parasitol* 2006; **100**: 703–14.
4. Smego RA Jr, Sebanego P. Treatment options for hepatic cystic echinococcosis. *Int J Infect Dis* 2005; **9**: 69–76.
5. Maravilla P, Andrew Thompson RC, Palacios-Ruiz JA, Estcourt A, Ramírez-Solís E, Mondragón-de-la-Peña C et al. *Echinococcus granulosus* cattle strain identification in an autochthonous case of cystic echinococcosis in central Mexico. *Acta Trop* 2004; **92**: 231–6.
6. McManus DP, Zhang W, Li J, Bartley PB. Echinococcosis. *Lancet* 2003; **362**: 1295–304.
7. Brunetti E, Junghans T. Update on cystic hydatid disease. *Curr Opin Infect Dis* 2009; **22**: 497–502.
8. Scherer K, Gupta N, Caine WP, Panda M. Differential diagnosis and management of a recurrent hepatic cyst: a case report and review of literature. *J Gen Intern Med* 2009; **24**: 1161–5.
9. Nelson JL, LoVecchio F. Echinococcosis presenting as acute abdominal pain. *Am J Emerg Med* 2010; **28**: 748.e1–e3.
10. Steta J, Torre A. Mexican-native human echinococcosis: case report of an underestimated disease. *Ann Hepatol* 2009; **8**: 251–4.
11. Méndez-Sánchez N, Vega R, Cortés R, Corona R, Pichardo-Bahena R, Uribe M. Hepatic *Echinococcus granulosus*: a case report. *Ann Hepatol* 2003; **2**: 99–100.
12. Palacios-Ruiz JA, Ramírez-Solís E, Moreno-Möller M, Cárdenas-Mejía A, Aguirre-Alcántara MT, Bonilla-Rodríguez C et al. [Identification and integral management of a case of autochthonous hydatidosis in Mexico]. *Rev Gastroenterol Mex* 2003; **68**: 41–5.
13. Mata-Miranda P, Osnaya-Palma I, Rodríguez-Prado U, Gutiérrez-Marín A, Tawil M, Hernández-González S et al. Epidemiologic and ultrasonographic study of echinococcosis in a community in the State of Mexico. *Am J Trop Med Hyg* 2007; **77**: 500–3.
14. Eckert J, Deplazes P. Biological, epidemiological, and clinical aspects of echinococcosis, a zoonosis of increasing concern. *Clin Microbiol Rev* 2004; **17**: 107–35.
15. Dziri C, Haouet K, Fingerhut A, Zaouche A. Management of cystic echinococcosis complications and dissemination: where is the evidence? *World J Surg* 2009; **33**: 1266–73.
16. Craig PS, McManus DP, Lightowlers MW, Chabalgoity JA, García HH, Gavidia CM et al. Prevention and control of cystic echinococcosis. *Lancet Infect Dis* 2007; **7**: 385–94.
17. Mortelé KJ, Segatto E, Ros PR. The infected liver: radiologic-pathologic correlation. *Radiographics* 2004; **24**: 937–55.
18. Junghans T, da Silva AM, Horton J, Chiodini PL, Brunetti E. Clinical management of cystic echinococcosis: state of the art, problems, and perspectives. *Am J Trop Med Hyg* 2008; **79**: 301–11.