Pelvic and retroperitoneal hydatid cysts superinfected with Brucella sp. and review of infected hydatid cysts

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Received 25 July 2012; received in revised form 27 December 2012; accepted 30 December 2012

Abstract. Hydatid disease is a zoonotic infection resulting from the tissue infestation of the larval stage of the parasite Echinococcus granulosus. Hydatid cysts superinfected with pyogenic organisms have been reported previously. Brucellosis is more prevalent in people with close contact to animals and those consuming fresh milk or fresh milk products. Although these two disorders have some similar epidemiological features, we did not encounter any hydatid cyst cases superinfected with Brucella species (sp.) in a search of medical literature (Pubmed). Here, we present a case of hydatid cyst disease superinfected with Brucella and review the literature on other hydatid cyst cases superinfected with pyogenic organisms. We conclude that in regions where brucellosis and hydatid cysts are endemic, cysts may be infected with Brucella sp.

INTRODUCTION

Hydatid disease is a zoonotic infection that results from the tissue infestation of the larval stage of the parasite Echinococcus granulosus. The life cycle of Echinococcus includes a definitive host (usually dogs) and an intermediate host (such as sheep, goats or swine). Humans are incidental hosts and represent usually the dead end for the parasite. Echinococcus granulosus adult tapeworms are usually found in dogs or other canids. On ingestion of E. granulosus eggs, hydatid cysts are formed mostly in liver and lungs, and occasionally in other organs, which are considered as uncommon sites of localization of hydatid cysts (Mandal & Mandal, 2012). Hydatid cyst cases superinfected with pyogenic organisms have been reported previously (Ferran et al., 1986; Blenkham et al., 1987; Masterton et al., 1987; Sitaram et al., 1990; Baykal & Belgin, 1997; Agarwal et al., 2000; Chen et al., 2002; Aslam et al., 2005; Turkgoglu et al., 2005; Kismet et al., 2006). Brucellosis is a common zoonotic infection with multisystemic involvement caused by Brucella species. Transmission of it to humans occurs mainly through the consumption of infected, unpasteurized animal milk and milk products. It is prevalent in our country and epidemiology of hydatid disease and brucellosis may overlap in some parts. (Kokoglu et al., 2006). We did not encounter any hydatid cyst cases superinfected with Brucella species (sp.) in a search of English medical literature (Pubmed, 1964 – June 2012). Here, we present a case of hydatid cyst disease superinfected with Brucella and review the literature on other hydatid cyst cases superinfected with pyogenic organisms.

Case

A 42 year-old male farmer was referred to our unit with intraabdominal cystic lesions. He presented to another hospital with complaints of bilateral leg pain, fever, myalgia and night sweats nine months before.
Initial tests revealed a normal complete blood count, erythrocyte sedimentation rate (ESR) of 70 mm/hour and C-reactive protein (CRP) of 93 mg/dL (normal < 5 mg/dL). He was given analgesics. Two months after, he developed swelling in the right testis, a rash on the abdomen and legs, pain and painful swelling in his left groin. *Brucella* standard tube agglutination test was positive (Wright: >1/160) and patient was given doxycycline and rifampin treatment. At that time, an abdomino-pelvic ultrasonography detected multiple cystic lesions (64 x 64 mm the largest) at the lower left quadrant and pelvis, as well as a hypoechoic lesion (50 x 29 mm) in the left psoas muscle and multiple necrotic lymphadenopathies in the left inguinal area. A contrast-enhanced abdominal computed tomography (CT) confirmed these lesions.

On admission, the results of the tests conducted in our unit were as follows: Hct 40%, Hb 13 g/dL, leukocytes 8900 /mm³, platelets 373,000 /mm³, ALT 12 U/L, AST 9 U/L, total protein 7.7 g/dL, albumin 3.2 g/dL, CRP 60 mg/dL; ESH 103 mm/h, Rose-Bengal’s test positive and Wright’s test positive (1/160; +++) A fine-needle aspiration biopsy sample (mud-colored cloudy abscess) was taken from the left inguinal area. Gram staining revealed gram-negative bacilli and *Brucella melitensis* were grown on solid media. Tuberculosis and fungal cultures of biopsy material remained sterile. Pathologic examination of the biopsy material revealed cuticular membrane remnants consistent with presence of hydatid cyst disease. Indirect hemagglutination test for *E. granulosus* was positive (1/512 dilution). Albendazole was added to the treatment. Cysts were removed by exploratory laparotomy. The patient followed monthly by routine biochemistry (especially transaminases for hepatotoxicity), CRP, ESR measurement with abdomino-pelvic ultrasonography. Treatment was continued for 6 months with albendazole (800 mg/day in 2 divided doses), doxycycline (200 mg/day in 2 divided doses) and rifampin (600 mg/day). After one year treatment completed, the patient had no additional complication.

DISCUSSION

*Echinococcus* and *Brucella* infections continue to be a health problem in endemic regions. Turkey is located in an endemic area for hydatid cyst and brucellosis (Mert et al., 2003; Yazar et al., 2006). Hydatid cyst disease prevalence was reported to be high in people with close contact to dogs and in countries like Australia, New Zealand, Brazil, Argentina, Spain, South America, Iran, North and East Africa where sheep farming is common (McManus et al., 2003). Brucellosis is another common infection in animals and it is more prevalent in people with close contact to animals (veterinarians, farmers etc.) and those consuming fresh milk or fresh milk products.

Extrahepatic hydatid cysts were reported to involve lungs (10–15%), spleen (0.9–8.9%), kidneys (1–4%), pancreas (0.25–0.75%), brain, heart, ovum, bone and abdominal wall (Hamamci et al., 2004). Hydatid cyst cases may present with symptoms of pressure or rupture in the location of cyst, allergic reactions or complications related to secondary infections.

Hepatic hydatid cyst cases superinfected with varying microorganisms including *Escherichia coli*, *Enterococcus* sp., *Staphylococcus aureus*, *Klebsiella* sp., *Haemophilus influenza*, and streptococci were reported in the literature (Table 1). In terms of extrahepatic hydatid cases, *Aspergillus fumigatus*, *Salmonella* sp., and streptococcal infected cysts in thorax (Agarwal et al., 2000; Aslam et al., 2005; García et al., 2010), a *Clostridium ramosum* infected case with brain involvement (Turkoglu et al., 2005), and a case of hydatid cyst with spleen involvement superinfected after cutaneous fistula formation (Kismet et al., 2006) were reported in the literature. Our literature search did not yield any previously reported cases of hydatid cyst with *Brucella* superinfection.

In uncomplicated cases of hydatid disease, the cyst content is clear as spring water, while infected cysts contain a purulent fluid. In the report of Chen et al. (2002), the
Table 1. A review of the location of the hydatid cysts and infecting microorganisms reported in the literature (References: Agarwal et al., 2000; Aslam et al., 2005; Baykal&Belgin et al., 1997; Blenkharn et al., 1987; Chen et al., 2002; Ferran et al., 1986; Garcia et al., 2010; Masterton et al., 1987; Sitaram et al., 1990; Turkoglu et al., 2005)

<table>
<thead>
<tr>
<th>The Affected Organ</th>
<th>Thorax</th>
<th>Liver</th>
<th>Brain</th>
<th>Retroperitoneum and Pelvis</th>
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<tr>
<td>Microorganism</td>
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<td>(Number of cases)</td>
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<tr>
<td><em>Salmonella</em> sp. (2)</td>
<td><em>Salmonella</em> sp. (2)</td>
<td><em>Clostridium ramosum</em> (1)</td>
<td><em>Brucella</em> sp. (1)*)</td>
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<tr>
<td><em>Streptococci</em> (2)</td>
<td><em>Streptococci</em> (4)</td>
<td><em>Yersinia enterocolitica</em> (1)</td>
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<td><em>Aspergillus</em></td>
<td><em>Klebsiella</em> sp. (2)</td>
<td><em>Haemophilus influenzae</em> (2)</td>
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<td><em>fumigatus</em> (3)</td>
<td><em>Yersinia</em> enterocolitica (3)</td>
<td><em>Escherichia</em> coli (4)</td>
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<td></td>
<td><em>Klebsiella</em> sp. (3)</td>
<td><em>Aeromonas hydrophila</em> (1)</td>
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<td></td>
<td><em>Acinetobacter baumannii</em> (1)</td>
<td><em>Pseudomonas aeruginosa</em> (1)</td>
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<td></td>
<td><em>Enterococcus</em> sp. (3)</td>
<td><em>Staphylococcus aureus</em> (2)</td>
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*A hydatid cyst in spleen superinfected after cutaneous fistula formation was reported but the microorganism was not defined (Kismet et al., 2006)

**Present report

Figure 1. Contrast-enhanced abdominal CT. Multiple lesions of varying sizes (largest one is approximately 6 x 7 cm) are found in left paraaortic and paraaortic areas

cyst was removed by laparotomy and short-term albendazole treatment was applied together with cefazolin + gentamicin combination against *Klebsiella pneumoniae* infection, resulting in successful cure. Agarwal et al. (2000) used PAIR (Puncture Aspiration Injection Reaspiration) treatment, but noticed an increase in cyst size accompanied by fever. Culture of drainage fluid yielded *Yersinia enterocolitica* serotype 3 and they reported cure with a 6 week course of albendazole + amikacin treatment
(Agarwal et al., 2000). Kismet et al. (2006) reported an infected splenic cyst with cutaneous fistula, but the pathogen could not be isolated. Cure was obtained by partial cystectomy, drainage and albendazole treatment. Turkoglu et al. (2005) reported a hydatid cyst case with brain involvement infected after bacteremia due to an intervention for pleural empyema. Following a large craniotomy and cortical incision, C. ramosum was isolated from the purulent cyst content. Cure was obtained with a 4 week course of albendazole + meropenem treatment.

In the present case we suspected an intraabdominal abscess because the case had not improved despite an appropriate treatment for brucellosis and a painful swelling had developed in the left inguinal area. Microbiologic and pathologic examination of the fine-needle aspiration biopsy sample obtained from this abscess revealed a hydatid cyst infected with Brucella sp. Pelvic cysts of the patient were removed by laparotomy and a large volume of purulent fluid was drained. Some small cysts could not be removed due to difficult anatomical position. Patient was followed with 6 courses of albendazole, doxycycline and rifampicin.

Secondary bacterial infection is a rare complication of hydatid cysts, occurring in 5-8% of cases, mostly as a result of pericystic and endocystic ruptures (Marti-Bonmati et al., 1990; Pedrosa et al., 2000). In a recent cohort of hydatid cysts, Garcia et al. (2010), described that 7.3% of 503 patients had a super-infected cyst. Cases of cyst infections occurring via hematogenous route without rupture have also been reported. The cyst anatomy is composed of three layers. The outside layer called pericyst (ectocyst) is derived from host tissue. This layer is composed of thin fibrous tissue and it is anatomically not possible to separate it from the host. Pericyst provides mechanical support for the cyst and allows the exchange of nutrients through its capillaries. Inside, a 1-2 mm layer called laminar membrane gives the cyst its spherical shape. Finally, a very thin and transparent germinative membrane constitutes the innermost layer where scolices are produced and released into the cyst fluid (Sherlock & Dooley, 1997). Wasunna et al. (1991) suggested that albendazole treatment may increase permeability in the laminar membrane and thus allow for bacterial transfer. However, in most cases of secondary infection albendazole was not previously used. The pericystic (adventitial) layer calcifies with time and vascular structure allowing feeding from the host is lost. None of the hydatid cyst cases with secondary infection (including our case) showed signs of calcification as detected by imaging studies. Hence, in the absence of calcification, secondary infection risk may be present following bacteremia.

In conclusion, in regions where brucellosis and hydatid cysts are endemic, cysts may be infected with Brucella sp. This is the first multivesical hydatid cyst case with retroperitoneal and pelvic involvement infected with Brucella sp. in the literature. Purulent cyst fluid present in all infected cases should be a warning sign for surgeons performing hydatid cyst surgery and cyst content should be subjected to microbiologic examination in addition to pathological assessment. Asymptomatic cases followed without surgical treatment should be warned regarding the possibility of secondary infections, and cyst infection should be considered in cases presenting with symptoms of localized infection.

REFERENCES


