Abdominal echinococcosis during pregnancy: clinical aspects and management of a series of cases in Chile

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SUMMARY Twelve pregnant women with hydatid disease are presented with median age of 29; 11 (91.7%) had a liver cyst and one (8.3%) had a kidney cyst as the primary disease location. Four (33.3%) had additional cysts located in the pelvis, peritoneal cavity and/or spleen; eight (66.7%) had two or more abdominal cysts. Three patients (25.0%) had surgery at the 3rd month after delivery and nine (75.0%) during their pregnancy. There was no histological evidence of hydatid disease in placentas, and no serological evidence of echinococcosis in the newborns was confirmed. One patient died after surgery. After a mean follow-up time of 39.5 months, we found one recurrent case of pelvic hydatid disease. Management of abdominal echinococcosis during pregnancy is an uncommon and difficult problem owing to the serious potential risks for mother and child.

Introduction

Hydatid disease is an important zoonosis in Chile, especially in the 9th region (39° south), with a prevalence rate of 18 to 48 cases per 100 000 inhabitants[1]. The high prevalence and incidence rate gives rise to a variety of uncommon disease presentations such as hydatid disease during pregnancy.

Because abdominal echinococcosis is typically non-symptomatic[2] and digestive symptoms are frequent during pregnancy, the diagnosis of abdominal echinococcosis would often be missed, mainly in areas with elevated prevalence and high grade of suspected diagnosis. Therefore, the diagnosis can often be established by routine abdominal ultrasound in prenatal control[3,4].

The natural history of echinococcosis in pregnant women is not well understood due to its low prevalence rate. In addition, there are very few cases reported in the literature and related knowledge is scarce[3–11]. In 1997, we reported our first experience in this field and since then, we have developed a protocol for the management of these patients[12].

The aim of this study is to present our experience in the management of abdominal echinococcosis diagnosed during pregnancy.

Patients and methods

We conducted a descriptive study of incident and consecutive cases at the Surgery and Obstetrics Departments of the Regional Hospital of Temuco, Chile. All pregnant women in different stages of pregnancy with coexistent abdominal hydatid cyst referred to our hospital between January 1997 and March 2000 were included. No exclusion criteria were considered.

The diagnosis was suspected by abdominal ultrasound findings in routine prenatal control, and confirmed by specific serological tests (enzyme-linked immunoassay–IgG and IgE). Moreover, thoracic X-ray was done to rule out lung echinococcosis[2,13]. Patients were treated with early surgery once the diagnosis was made by using the following cyst aspects for therapy decision: location, size, number and evidence of complications, and gestational age. Related to location, we believe that all hydatid cysts of pelvic and peritoneal location require surgery prior to delivery. We considered hepatic cysts located at segments III, IV, V, and VI to have higher risk of rupture; as well as those located in other abdominal organs which had grown into the major and/or minor pelvis. In relation to cyst size, we believe that all those greater than 5 cm in diameter require surgery prior to delivery. With respect to the presence of cyst complications, we consider that the following aspects must be treated immediately: imminent rupture, biliary tract communication, infection, and hepatothoracic transit because of potential high risk of morbidity in mother and/or child.

However, if surgery is indicated for the patient, and diagnosis is confirmed early in her pregnancy, surgery has to be performed during her first trimester. If diagnosis is made during the second trimester, surgery has to be performed before pregnancy ends. If the diagnosis is made during the third trimester or at the end of pregnancy, surgery has to be deferred, in general, to the puerperium. Whenever there is vital risk to mother or child, surgery should take place immediately if the chance of eventual complications during delivery is coming from cyst infection, hepatothoracic transit or anaphylactic shock[14]. Regarding pregnancy resolution, in these cases, we prefer normal delivery, but with patients with large abdominal cysts which have been diagnosed at the end of pregnancy we choose elective caesarean delivery. Complementary therapy with albendazol is used only in patients with multiple hydatid cyst disease.

Placentas were histologically studied to discard placental compromise. In addition, all newborns were studied with specific serological tests at delivery and at the third and sixth month of life to identify unexpected child infection with echinococcosis.

This protocol was submitted and approved by the Ethics Committee of the Hospital in agreement with the Helsinki Declaration.

Exploratory data analysis and summary measures were used for the statistical analysis.

Results

Twelve pregnant women were included in the study. All patients had abdominal hydatid disease. Their median age was 29 (21 to 41 years). Eleven women (91.7%) had liver cyst location, and one (8.3%) kidney cyst as the primary lesion. Four patients (33.3%) had, in addition, pelvis lesions (bladder, Douglas cul-de-sac, ovarian or anterior uterus surface [Figure 1]), peritoneal cysts (mesentery and

omentum) and/or spleen locations; and 8 (66.7%), had two or more abdominal cysts (Table 1).

Surgery was performed on three patients (25.0%) after the third month post delivery, the other nine in pregnancy: one during their first trimester (week 14), six during their second trimester (weeks 18, 20, 23, 24, 26 and 27) and two in the third trimester and at delivery (weeks 30 and 38, respectively). Median surgical time was 220 min (95 to 450 min). The mean postoperative stay was 8 days (2 to 22 days), and no evidence of anaphylaxis was detected in the patients. Total cystectomy (in peritoneal, most of the pelvic location and some of the liver cysts), partial pericystectomy in kidney, spleen and some liver location (no splenectomy was necessary), and total pericystectomy in a few liver cysts were performed.

Nine patients (75%) underwent normal delivery and three (25%) needed caesarean operation. There was no histological evidence of hydatid disease in placentas, and in the newborns no serological evidence of echinococcosis was confirmed in both measurements.

In four patients with multiple hydatid cyst disease, albendazol was used for 3 months after suspension of breast-feeding. Nevertheless, during follow-up (median time 40 months [22 to 68 months]) one case of multiple pelvic hydatid recurrence was confirmed.

Two patients had surgical morbidity: one had pulmonary atelectasis and treatment was respiratory exercises; the other had superficial surgical site infection and treatment was simple wound care. One 33-year-old patient with multiple liver cysts (one in transit to the thorax and the other infected) died due to massive pulmonary thromboembolism during the third day after surgery.

Discussion

A total of 5000 deliveries per year are attended in the Regional Hospital of Temuco (reference centre for the region). In general, pregnant women have at least three routine prenatal controls, two of which include abdominal ultrasound exploration. The estimated prevalence of echinococcosis in pregnant women in the region (for the past 3 years) is 0.07%.

In the literature there are few reports and poor information about this health problem[3,4,8,10,15,18]. In our experience the most frequent cyst localization is the liver (in both men and non-pregnant women), and there is a high risk of rupture to the peritoneal cavity causing anaphylaxis and cyst spread to peritoneum, abdominal organs and pelvic structures[8,18]. This is especially relevant in pregnant women for the alteration of uterine growth and subsequent problems during delivery[7,8].

Regarding diagnosis it is important to emphasize the role of epidemiological risk factors such as living in rural areas and in some specific mountain and coastal cities (where the highest prevalence rate occurs in this region)[1,13]. Although specific immunoglobulin determination is important[1] abdominal ultrasound is the gold standard, as it permits pregnancy control and shows the cysts, their number, location and relationship with other abdominal structures[2].

Other important issues are anaphylaxis and cyst infection[2,5], premature delivery[8], dystocic dynamic[7], dystocic obstructive and uterine rupture[15]. The likelihood of fetal infection through the placenta has been discussed by other groups[4,17], but association has not been proven. There is some incipient evidence of maternal transfer protection from echinococcosis in animals[19], however in this series we did experience this complication.

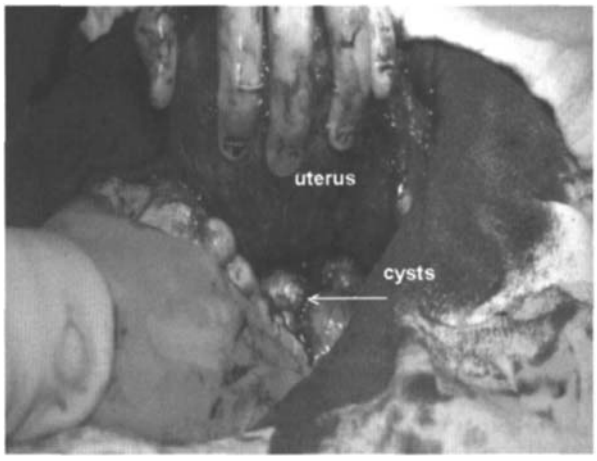

Fig. 1 Multiple pelvic hydatid cysts surround gravid uterus in a pregnant woman of 38 weeks' gestation

The treatment of hydatid disease is mainly surgical, although during pregnancy other options can be considered:

(1) Observation: we do not regard this option as a routine approach as cyst complications appearing during pregnancy may be fatal for both mother and/or the fetus. The exceptions are those cases in which small cysts are located in posterior liver segments, upper abdominal organs and retroperitoneum; in such cases complications are unlikely to take place.
(2) Simple aspiration guided by ultrasonography[16,17]: we think this alternative has great risks for anaphylaxis and peritoneal dissemination[20]; in fact, some experience of this has been published elsewhere[21].
(3) Medical treatment: albendazol and mebendazol have teratogenic effects in animals[22], thus their use must be contraindicated in pregnant women[23].

The low morbidity in this series is similar to other studies[3,4,18] but there are no reports of mortality in those

Table 1 Characteristics of hydatid disease patients (n=12)

Characteristic	*No. cases*	*Percentage*
Location of lesions*		
Liver†	11	91.7
Kidney†	1	8.3
Peritoneum	4	33.3
Spleen	2	16.7
Pelvis	2	16.7
Number of lesions per patient		
One	4	33.3
Two	2	16.7
Three or more	6	50.0
Complications‡		
Without complications	9	75.0
Cyst infection	3	25.0
Thoracic transit	1	8.3
Characteristic	Median	Minimum–maximum
Largest cyst size (cm)§	15.0	(6.0–25.0)
Extirpated cyst weight (kg)¶	7.0	(0.1–14.0)

*Two of the patients had pelvic and peritoneal location associated with liver cysts, and one of these also had a spleen cyst
†Primary cyst location. The others are secondary locations
‡One of the patients had cyst infection and thoracic transit
§Size of the greatest lesion
¶Weight of extirpated lesions

groups. With respect to the patient who died, it is important to point out that she had four liver cysts, one of them was infected (transformed in liver abscess), and another 25-cm-diameter cyst was in transit to the thorax with high risk of rupture. Surgery was indicated for this patient based on severe dyspnoea, sepsis and risk of cyst rupture with secondary anaphylaxis.

In conclusion, the problems of hydatid disease in pregnancy are related to the difficulties of detecting it (because of its non-symptomatic course), the different risks of cyst complications, the potential alteration of uterine growth and subsequent problems during delivery, and the complexity of surgical procedures to treat it.

References

1 Gutiérrez R, Muñoz F, Oberg C, Ampuero F. Conclusiones de las II Jornadas nacionales de hidatidología. [II National meeting of hidatology conclusions.] *Rev Méd Chile* 1991;**16**:8–9

2 Manterola C, Acencio L, Garrido L, Bahamóndez JC, Barroso M. Hidatidosis hepática. Estudio descriptivo de aspectos clínicos y terapéuticos de una serie consecutiva de casos. [Hepatic hydatid disease. Descriptive study of clinical and therapeutics aspects of a consecutive series.] *Rev Chil Cirugía* 1997;**49**:352–9

3 Baba A, Chaieb A, Khairi H, Keskes J. Profil épidemiologique de l'hydatidose pelvienne. A propos de 15 cas. [Pelvic hydatid disease epidemiological profile of 15 cases.] *J Gynecol Obstet Biol Reprod* 1991;**20**:657–60

4 Kekih MA, Abed H, Cheli M. Kyste hydatique pelvien et grossesse. A propos de 4 cas. [Pelvic hydatid cyst. Report of 4 cases.] *J Gynecol Obstet Biol Reprod* 1992;**21**:803–5

5 Horzic M, Bunoza D, Maric K. Anaphylactic shock in a female patient due to spontaneous rupture of a hepatic hydatid cyst: a case report. *Hepatogastroenterology* 1996;**43**:1601–2

6 Semchyshy S. Echinococcus discovered during pregnancy. *Am J Obstet Gynecol* 1970;**107**:477–83

7 Bickers VM. Hydatid disease of female pelvis. *Am J Obstet Gynecol* 1970;**107**:477–83

8 Rahman MS, Rahaman J, Lysikibwicz A. Obstetric and gynaecological presentation of hydatid disease. *Br J Obstet Gynaecol* 1982;**89**:665–70

9 Blochle C, Lloyd DM, Izbicki JR, Schroder S, Brolsch CE. Right-sided hemihepatectomy in echinococcosis of the liver in pregnancy. *Chirurg* 1993;**64**:580–3

10 Golaszewski T, Susani M, Golaszewski S, Sliutz G, Bischof G, Auer H. A large hydatid cyst of the liver in pregnancy. *Arch Gynecol Obstet* 1995;**256**:43–7

11 Van Vliet W, Scheele F, Sibinga-Mulder L, Dekker GA. Echinococcosis of the liver during pregnancy. *Int J Gynaecol Obstet* 1995;**49**:323–4

12 Manterola C, Espinoza R, Barroso M, *et al.* Hidatidosis y embarazo. [Hydatidosis and pregnancy.] *Rev Chil Obstet Ginecol* 1997;**62**:33–7

13 Manterola C, Molina E, Fernández O, Barroso M. Quistectomía subtotal. Una alternativa quirúrgica en el tratamiento de la hidatidosis hepática. [Partial pericystectomy. A surgical alternative for liver echinococcosis treatment.] *Rev Chil Cirugía* 1998;**50**:621–9

14 Manterola C, Barroso M, Oberg C, Molina E, Vial M, Fernández O. Alternativas quirúrgica en el tratamiento de la hidatidosis hepática. [Surgical alternatives in the treatment of hepatic hydatidosis.] *Bol Chil Parasitol* 1999;**54**:13–20

15 Rahman MS. Dystocia due to hydatid disease of pelvis. *Garyounis Med J* 1978;1:6–10

16 Kain KC, Keystone JS. Recurrent hydatid disease during pregnancy. *Am J Obstet Gynecol* 1988;**159**:1216–17

17 Kidess EA, Akiel AS, Baquel HS, Malaika S. Echinococcosis: an obstetric and gynaecological view. *Ann Saudi Med* 1988;**8**:202–5

18 Ladeb MF, Bouahouala H, Slim K, Ganouni A. L'hydatidose pelvienne chez la femme. A propos de 3 cas. [Pelvic hydatid disease in women. Report of 3 cases.] *J Gynecol Obstet Biol Reprod* 1989;**18**:493–5

19 Dempster RP, Harrison GB. Maternal transfer of protection from echinococcus granulosus infection in sheep. *Res Vet Sci* 1995;**58**:197–202

20 Stoianov G, Grigorov N, Slavov V, Bogusheva E. Intraoperative anaphylactic shock following the percutaneous puncture of hepatic echinococcal cyst. *Khirurgiia (Sofiia)* 1995;**48**:12–13

21 Sánchez-Tirado JA, Carrión-Pareja JC, Lafuente-Martín FJ, Martínez-Ubieto J, Laglera-Trebol S. Comentarios del tratamiento del choque anafiláctico causado por rotura de quiste hidatídico hepático [Comments on the management of anaphylactic shock caused by rupture of an unsuspected hepatic hydatid cyst.] *Rev Esp Anestesiol Reanim* 1998;**45**:305–6

22 Mantovani A, Ricxciardi C, Stazi AV, Macri C. Effects observed on gestational day 13 in rat embryos exposed to albendazole. *Reprod Toxicol* 1995;**9**:265–73

23 Auer H, Kollaritsch H, Juptner J, Aspock H. Albendazol and pregnancy. *Appl Parasitol* 1994;**35**:146–7

Uterine fibroid in a man: how African tradition solved a social crisis

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Case history

A 70-year-old Zimbabwean tailor was referred with an intermittently palpable non-tender mass in the lower abdomen. The patient appeared to be an otherwise fit and healthy man with no previous medical history. Routine blood investigations revealed no abnormalities. An abdominal ultrasound scan was unable to elucidate the origin of the mass. Laparotomy was undertaken and the finding, to our surprise, was a fibroid uterus. The dimensions of the uterus were 70 × 60 × 45 mm and the largest fibroid at the fundus measured 160 × 110 × 100 mm. Both ovaries and fallopian tubes were present and unremarkable. This immediately led to an examination of the genitalia, which had been reported by the junior medical officer as 'normal'. In fact there were absent scrotum and testes, a phallus and apparent coronal hypospadias without chordee. Total hysterectomy with bilateral salpingo-oopherectomy was carried out; the patient made an uneventful recovery.

Histopathology confirmed a leiomyoma of the uterus with structurally normal ovaries and fallopian tubes. Post-operative blood investigations showed: progesterone 9.03 ng/mL; testosterone 3.66 ng/mL (normal 1.81–7.58);