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Uncommon Sites of Hydatid Disease

J. Prousalidis, M.D., K. Tzardinoglou, M.D., L. Sgouradis, M.D., C. Katsohis, M.D., H. Aletras, M.D.

First Propedeutic Surgical Clinic, Aristotelian University, AHEPA Hospital, St. Kyriakidi 1, 546 36 Thessaloniki, Greece

Abstract. Echinococcosis remains an endemic surgical problem in many Mediterranean countries. We report our experience with such cases when the disease is located in uncommon sites, outside the liver and lungs. This study was an effort similar to a previous one but with more cases and additional information. Between 1967 and 1994 a total of 49 patients suffering from hydatid cysts located in various organs other than the liver and lungs presented to our clinic. There were 28 men and 21 women, with their ages ranging from 10 to 66 years and 22 to 80 years, respectively. Among these patients, 25 had the parasitic cyst in the peritoneal cavity, 10 in the spleen, 5 in the kidney, 3 in the spinal column, 2 in the retroperitoneal space, 1 in the abdominal wall, 1 in the myocardium, 1 in the thoracic wall, and 1 in the thigh. Their hospital stay was 9 to 88 days (average 27 days). Only two patients—one with cardiac hydatidosis and one with spinal hydatidosis—died postoperatively. Three patients with multiple cysts in the peritoneum and one with cysts in the thigh had recurrences of the disease and were reoperated successfully.

Echinococcosis, usually found in the liver and lungs, can develop in the abdominal or pelvic cavity and more rarely anywhere in the body [1]. In this study a series of 49 patients with echinococcosis located at uncommon sites is reported, and the relative diagnostic and therapeutic problems are reported.

Materials and Methods

In the 1st Propedeutic Surgical Clinic of AHEPA Hospital, University of Thessaloniki, from 1967 to 1994, a total of 540 patients were admitted for hydatosis, 49 of whom were suffering from cystic echinococcus at various sites of the body other than the liver and lungs (Table 1). There were 28 men and 21 women, with ages ranging from 10 to 66 years (average 45 years) for the men and 22 to 80 (average 51 years) for the women.

Among the study group 14 patients had multiple cysts; 14 peritoneal cysts coexisted with hepatic cysts, one splenic cyst with a cyst of the liver, one splenic cyst with cysts of the liver and lung, one renal cyst with a hepatic cyst, and one retroperitoneal cyst with small peritoneal cysts.

The diagnosis in general was based on the history, clinical picture, and various laboratory and radiologic examinations (Table 2). The surgical procedure employed was planned according to the location of the cysts (Table 3).

Correspondence to: J. Prousalidis, M.D.

Peritoneum

One patient with peritoneal and liver cysts had previously undergone removal of peritoneal cysts, two patients with single peritoneal cysts had had removal of liver cysts, and one patient also with a peritoneal cyst underwent removal of liver and lung cysts. The symptomatology was generally indistinguishable from that of cysts of the liver. The most frequent presenting symptom of the 25 patients with cysts in the peritoneal cavity was abdominal pain and discomfort (12 cases); some gave a history of weight loss (3 cases), nausea, vomiting (3 cases), and dysuria (4 cases). In three patients the disease was almost asymptomatic and it was discovered by ultrasonic examination. Hydatid thrill was elicited in four instances. Casoni's intradermal test was carried out in 17 patients (14 positive results), a complement fixation test (CFT) in 17 (9 positive results), an indirect hemagglutination test (IHA) in 5 (4 positive results), and an eosinophil count (16 positive results). Ultrasonography and computed tomography (CT) of the abdomen were performed in six and seven cases, respectively, and showed accurate pictures of the cystic lesion (Fig. 1).

In the group with peritoneal cysts the surgical incision, double subcostal or vertical, was large enough to permit thorough examination of the entire abdominal cavity and management of any coexistent cysts. It was possible to remove intact pediculated cysts in 13 cases; partial pericystectomy with parasite evacuation and abdominal drainage was performed in 5 patients; and in 5 both methods, in different cysts, were performed. The strategy chosen was based on the adhesions around the cysts, which created the danger of bleeding or rupture of the cysts during the dissection. In one case partial pericystectomy was accomplished by epiploplasty. In two patients, after removing a number of cysts it was necessary to terminate the operation, and some small cysts without tension were left behind. Another patient was too old to be operated, and it was decided to keep him under observations.

Spleen

In the 10 patients with splenic cysts, splenomegaly was the most frequent diagnostic sign (8 cases). In five cases calcification of the cyst was seen on the abdominal film, in four the Casoni's test was positive, in five Weinberg's complement fixation test was positive, and in four the sonographic and CT results were indicative of hydatosis of the spleen (Fig. 2).

Table 1. Uncommon sites of hydatid disease.

Location	No. of cases
Peritoneal cavity[a]	25
Spleen[b]	10
Kidney[c]	5
Retroperitoneal space[d]	2
Abdominal wall	1
Myocardium	1
Thoracic wall	1
Spinal column	3
Thigh	1
Total	49

[a]In 14 cases plus liver cysts.
[b]In one case plus liver cysts; in one case plus liver–lung cysts.
[c]In one case plus liver cysts.
[d]In one case plus small peritoneal cysts.

Table 2. Methods for diagnosing uncommon hydatid cysts.

Site and diagnostic test	No. of cases
Peritoneum	
Casoni	14
Complement-fixation test (CFT)	9
Indirect hemagglutination test (IHA)	4
Eosinophil count	16
Ultrasonography	6
Computed tomography (CT)	7
Spleen	
Casoni	4
CFT	5
Ultrasonography	4
CT	4
Retroperitoneum	
Intravenous pyelography (IVP)	2
Kidney	
CFT	2
IVP	2
Isotope scanning	2
Ultrasonography	2
CT	2
Spinal column	
Plain radiography	3
CT	3

Table 3. Diagnostic methods for uncommon hydatid cysts.

Site and diagnostic method	No. of cases
Peritoneum	
Casoni	14
CFT	9
IHA	4
Eosinophilia	16
Ultrasonography	6
CT	7
Spleen	
Casoni	4
CFT	5
Ultrasonography	4
CT	4
Retroperitoneum	
IVP	2
Kidney	
CFT	2
IVP	2
Isotope scanning	2
Ultrasonography	2
CT	2
Spinal column	
Plain radiography	3
CT	3

See Table 2 for abbreviations.

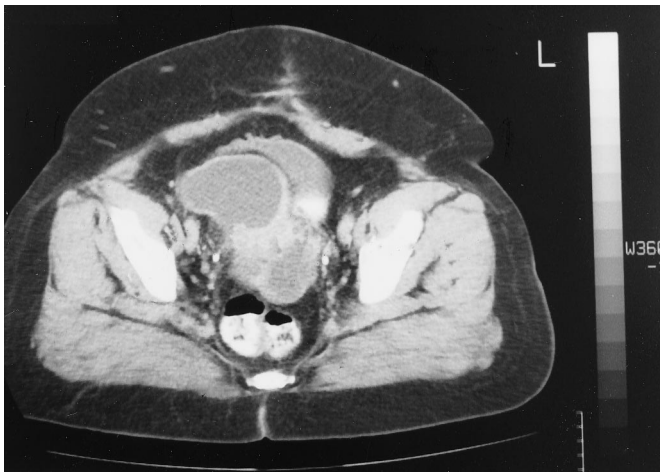

Fig. 1. Peritoneal cysts.

Seven patients with splenic cysts underwent splenectomy without technical difficulty or rupture of the parasite. One patient, 10 years old, also had cysts in his liver and left lung. The spleen and two cysts in the liver were removed through a right subcostal incision during the first procedure, and three cysts were removed from his left lung through a thoracotomy at a second operation. During laparotomy a 75-year-old patient with a large calcified cyst of the spleen was found to have neoplastic deposits in the liver, and a biopsy was done. Two patients refused operation.

Kidney

Among the five patients with renal cysts, one had undergone removal of a hydratic cyst of the lung 1 year before; because of the nonspecificity of the clinical picture and investigations, the diagnostic process generally was not easy. Fever, hematuria, dysuria, pain or discomfort in the loin (three cases), and a palpable mass resembling a renal tumor (two cases) were the symptoms and findings. For diagnosis of the parasite, a plain abdominal radiograph and intravenous pyelography (IVP) two cases each, showed the calcified cystic tumor compressing the kidney pelvis and parenchyma. The complement fixation test (two cases), isotope scanning (two cases), sonography (two cases), and CT (two cases) were especially helpful (Fig. 3).

After establishing the diagnosis of cysts in the kidney, the extent

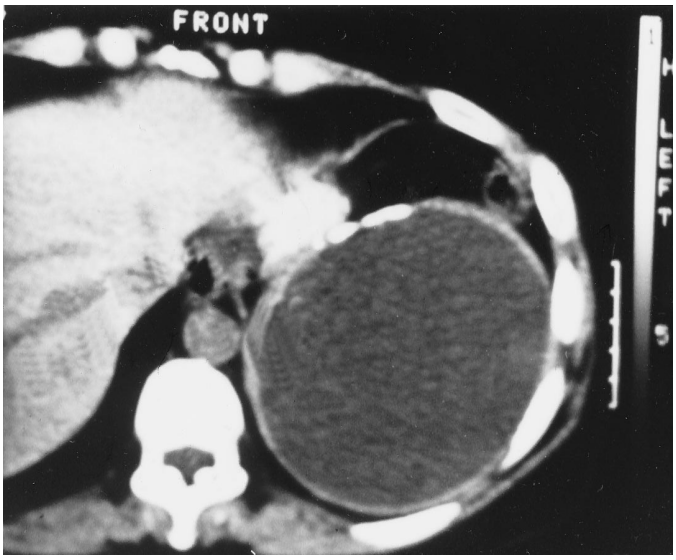

Fig. 2. Splenic cyst.

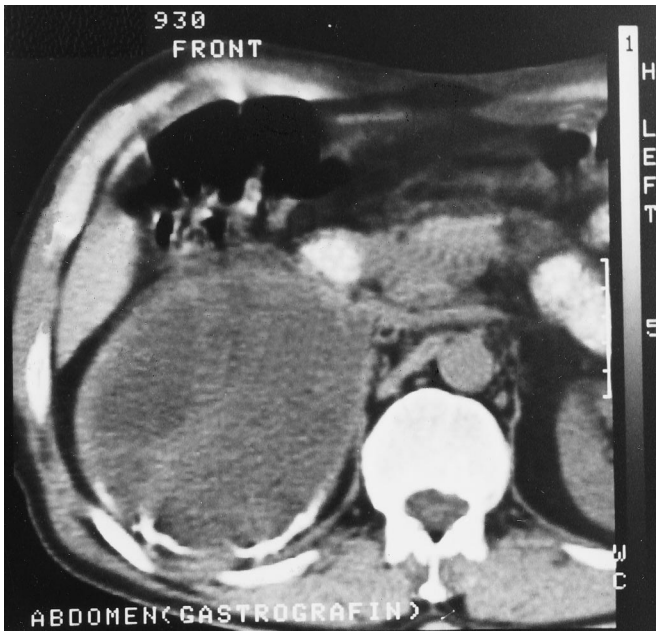

Fig. 3. Renal cyst.

of the excision depended on the therapy chosen, a decision made during the operation. In one case the kidney was so damaged nephrectomy was necessary. In two cases after removal of the parasite the kidney was left intact, and in two others after cautious evacuation of the cyst partial pericystectomy with drainage was undertaken, leaving no sizable opening into the pelvis.

Retroperitoneum

Clinical examination of the patients with retroperitoneal cyst revealed a palpable mass in the loin, extending down to the iliac fossa. The patients complained of local, dull pain. Pelvicalyceal distortion on IVP, performed in two cases, helped during diagnostic workup.

The cysts in the retroperitoneal space were associated with dense vascular adhesions to the surrounding abdominal organs. Therefore evacuation was chosen, with partial pericystectomy and drainage of the remaining cavity.

Abdominal Wall

Physical examination of patients with abdominal wall cysts clearly revealed a soft cystic mass in the subcostal incision for previous hydatid surgery. Dissecting the cyst from underneath the liver and large intestine was difficult, and the parasite was removed by partial pericystectomy and drainage.

Heart

The patient with myocardial cysts had been treated conservatively at the cardiac clinic 2 years before for "serous pericarditis." A month later he was operated on to remove two ruptured echinococcal cysts from the left ventricle. He was admitted later to our surgical clinic for "embolism" in his popliteal artery.

During the emergency "embolectomy" parts of a hydratid cyst were removed by use of a Fogarty catheter. The patient had been operated elsewhere for two cysts in the pericardium, and one in the brain and in our unit for another parasitic peripheral "embolization," respectively, 1, 4, and 5 years later. The peripheral arterial emboli were easily managed without perioperative coronary or cardiac disturbances.

Thoracic Wall

The patient with a cyst in the thoracic wall had been twice operated on for cysts of the posterior surface of his thorax, and he complained of an infected lump under the thoracic scar. Because of the coexistent adhesions total removal of the parasite was impossible, and evacuation of the cystic cavity and local drainage were undertaken.

Spinal Column

The three patients with disease in the skeletal system reported a long-lasting history of atypical "rheumatism." One of the patients with a hydatid cyst of the thoracic spine affecting the fifth and sixth thoracic vertebrae and another patient with a cyst of the lumbar spine affecting the second and the third lumbar vertebrae were admitted for paraplegia. The second of these patients was referred to the neurosurgical department. One patient with a cyst of the lumbar vertebrae was admitted because of an external fistula and secondary regional infection. The plain radiograph and CT scan were diagnostic not of the disease itself but of the destructive echinococcal process. In the patients with neurologic manifestations the CT scan showed cord compression as well. The final diagnosis was based on examination of the parasitic material that was removed during operation.

For the cyst of the thoracic spine, evacuation of the daughter cysts and relative curettage and drainage of the residual cavity

were done. Conservative treatment with local antibacterial agents and curettage was performed for the infected lumbar spine.

Thigh

The CT scans easily revealed hydatid disease in patients with cysts in the thigh. The patient with hydatosis of the thigh had twice—3 and 5 years previously—undergone removal of a hydatid cyst from the groin at the same site. In this case, eight infected cysts were found between the adductor muscles and were removed.

Results

All but two of the patients had an uneventful recovery. The patient with a second peripheral arterial embolism, although the embolectomy was successful, died from pneumonia on the 18th postoperative day. The patient with thoracic spine hydatidosis died 50 days after the operation due to cardiorespiratory failure. The postoperative hospitalization ranged from 9 to 88 days (average 27 days). This period was extended in some patients owing to a number of complications related to concomitant hydatid liver surgery (usually the drainage).

One patient initially operated for large retroperitoneal and small peritoneal cysts had recurrence of the peritoneal cysts 5, 7, and 10 years later. One patient with operated peritoneal and hepatic cysts developed hydatid cysts in the liver and then in the spleen, and tail of the pancreas 1 and 12 years later. In this case, in addition to management of the liver cysts, splenectomy for the splenic cysts and cystopericystectomy for the pancreatic cyst were performed. One patent operated initially for multiple peritoneal and liver cysts and after 3 years for a cyst in the lung, 22 years later developed hydatid cysts of the kidney, uterus, and liver. For the renal cyst, because of severe hydronephrosis and serious infections, nephrectomy was chosen. The uterine cyst was treated by partial pericystectomy and drainage. One patient with cysts in the thigh was reoperated 10 years later to remove cysts at the same site. During the follow-up period, 1 to 26 years after the initial operation, the others (as far as we know) had not been reoperated.

Discussion

There is no question that we generally have few diagnostic problems with *Echinococcus granulosus* [2]. Ultrasonography and CT easily detect the disease in patients. The other exception may be those with hydatid cysts at uncommon sites of the body. The surgeon must be aware of this possibility so as to follow the special therapeutic policy necessary and to avoid dangerous "spillage" of the parasite.

In the present series, except of some diagnostic difficulty with heart and bone cysts, generally the definitive diagnosis of the disease was easily established. It must be noted that sonography and CT were not available in our early cases. According to literature, the clinical picture, special laboratory tests (Casoni, CFT, IHA), the plain radiographs, displacement of the viscera seen by radiographic contrast studies (IVP) can provide the necessary information [2, 3]. Lastly, the newer organ-imaging methods, with their accurate pictures, have simplified the diagnostic process. The intradermal Casoni's test no longer has a place [2]. Serologic examinations have the problems of low diagnostic sensitivity and specificity and now have only limited use [2]. The indirect fluorescent antibody test performed by some laboratories gives a diagnostic result of 78% and remains positive for 2 to 3 years [2].

In addition to the common cysts of the liver and lung, those of the peritoneal cavity are quite frequent [1]. They account for 10% to 16% of cases and are mainly a result of rupture of concomitant liver cysts [3, 4]. Among our 540 cases with hydatid disease, those with peritoneal cysts accounted for 5% of cases. The primary variety of the disease, carried to the peritoneal cavity by the arterial circulation, is rare [5]. We assume that eight of our patients with peritoneal cysts and one with a retroperitoneal cyst have been infected via this route because, as we reported before, none had a history of or coexistent hydatid disease.

In the literature the spleen is rarely contaminated by *Echinococcus*, with a reported ratio of 3% of patients with hydatid disease. In our series the incidence of splenic cysts was 2%. There is the belief that the embryo may migrate to this location from the portal vein during bouts of increased abdominal pressure. We noted that six splenic cysts in our patients had developed via this mechanism. In the patient with a coexistent liver cyst, in the one with coexistent liver and lung cysts, and in one who later developed echinococcosis of the liver, spleen, and pancreas, we assumed the secondary nature of the cysts. Parasitic cysts of the pancreas are reported rarely [8].

Parasitic cysts of the kidney are rare, with an incidence in other series and ours of 2.5% and 1.0%, respectively [9]. Hydatid disease in these cases is usually of the primary type, with the organ receiving the hexacanth embryo through the arterial circulation [1]. Only in one of our patients with a concomitant liver cyst and in the reoperated patient with renal and uterine cysts was the parasite seeded from the existent peritoneal and hepatic hydatidosis.

The uncommon hydatid cysts of the heart, although they sound like a serious situation, can potentially be treated [10]. Cysts of the myocardium are considered primary, but those of the pericardium are secondary. One of the complications is peripheral emboli [1]. Our patient with an operated cyst of the myocardium and pericardium had been treated twice for emboli in the legs and once for emboli in the brain.

The parasitic cysts in the abdominal or thoracic incision and the cyst in the thigh were of the secondary type, easily ruptured and recurrent. The especially debilitating hydatid disease of the bones is of the primary type and can affect any part of the skeleton [11]. Differential diagnosis from other orthopedic diseases is difficult. The disease may have, as in two of our patients, neurologic sequelae and a generally poor prognosis. The parasite is usually controlled surgically, but the disease is not necessarily cured, as it is often complicated by secondary infection.

For treatment the general rules of hydatid surgery were followed [3, 4]. The cysts, when possible, were totally removed without sacrificing the organ(s) involved. This procedure was feasible in 14 cases with peritoneal cysts (13 operated initially, 1 three times later), two with renal cysts, and one with a cyst of pancreas. Whenever the cyst seems to be incorporated into the organ, the aim is to evacuate the contents of the cyst without spillage and to perform a partial pericystectomy. The residual cavity can be drained or filled with omentum. This policy was applied in five patients with peritoneal cysts, in two with renal cysts, in two with retroperitoneal cysts, in those with a uterine cyst,

and in those with cysts of the thoracic and abdominal wall and the thoracic column. Both methods were performed in five patients with peritoneal cysts. In the patients with cysts of the spleen and in two with cysts of the kidney (one initially, one operated later), removal of the organ was mandatory. The decision for nephrectomy, as reported by others and as we have seen in our series, is due to the large size of the cysts, serious damage to the organ, and obstruction with secondary infection [12]. Management of the patient with hydatid arterial mobilization was problematic. Although the cysts of the myocardium had been removed initially, there were later admissions for popliteal embolectomy, removal of pericardial cysts, brain emboli, and again for femoral emboli. This series of procedures was probably due to rupture of the parasite at the initial operation and subsequent relapse.

The outcome of surgery is good. In agreement with others, we had seen no postoperative mortality or severe morbidity among the cases with peritoneal cysts [13]. Both of the deaths in our series occurred in patients with severe types of uncommon hydatidosis: of the heart and bones, respectively. Although they were not directly attributed to the operation, they were propably related to it. The first patient died from pulmonary dysfunction after repetitive embolization and the second from immobilization and disability. The mean hospital stay, although extended, was not related to the treatment of the cysts reported here but to coexistent liver cysts. It was specifically prolonged in all the cases in which a drainage tube had to be applied after excising the wall of the cyst. The recurrence rate of peritoneal cysts noted elsewhere is 13%. Our rate in general was similar: 8.5%.

In conclusion, our results seem satisfactory. We hope that our analysis of the presentation, diagnosis, and treatment employed for uncommon hydatid cysts, in this and other series, will prove useful. Echinococcosis, with increasing migration, is a global problem; and every effort to improve its treatment is worthwhile [14]. With the collation of genetic, biologic, diagnostic, and prevalence data of all echinococcal forms, the future outlook for treating the disease is promising [15, 16].

Surgery is still the treatment of choice. As an alternative or supplementary nontoxic scolicidal agents or combination chemotherapy with puncture of the cysts, aspiration of fluid, introduction of protoscolicide, and aspiration probably must be tested [16–20]. As for some of our patients, candidates for chemotherapy are those at poor risk, those with disseminated hydatid disease, or those with possible secondary echinococcosis following rupture of a cyst.

Résumé

L'échinococcose reste essentiellement un problème chirurgical endémique dans beaucoup de pays Méditerranéens. Dans cet article nous rapportons notre expérience de l'échinococcose de sites anatomiques peu habituels, c'est à dire en dehors du foie et des poumons. Cet article complète un travail antérieur déjà publié, mais avec plus de cas. Dans la première clinique propédeutique chirurgicale entre 1967 et 1994, nous avons traité 49 patients ayant un kyste hydatique localisé en dehors du foie et des poumons. Il y avait 28 hommes et 21 femmes âgés, respectivement, de 19 à 66 et de 22 à 80 ans. Vingt-cinq patients avaient un kyste de la cavité péritonéale, 10 de la rate, cinq du rein, deux dans l'espace rétro-péritonéal, un dans la paroi abdominale, un du myocarde, un dans la paroi thoracique, trois au niveau de la colonne vertébrale et un dans la cuisse. La durée du séjour allait de 9 à 88 jours avec une moyenne de 27 jours. Deux patients, un ayant un kyste cardiaque et un autre ayant un kyste spinal, sont décédés. Trois patients avec des kystes péritonéaux et un avec un kyste de la cuisse ont récidivé et ont été réopérés avec succès.

Resumen

La equinococosis se mantiene como problema quirúrgico endémico en muchos países del Mediterráneo. En la presente comunicación estamos informando nuestra experiencia con casos de ubicación poco común de la enfermedad, con exclusión del hígado y los pulmones; se trata de un reporte similar a uno previamente publicado, pero con un mayor número de casos e información adicional. En el período entre 1974 y 1994, se trataron 49 pacientes con quistes hidáticos localizados en diversos órganos, diferentes del hígado y los pulmones, 28 hombres y 21 mujeres, con edades entre 19 y 66 años y 22 y 80 años, respectivamente. Veinticinco pacientes presentaron el quiste parasitario en la cavidad peritoneal, 10 en el bazo, 5 en el riñón, 2 en el espacio retroperitoneal, 1 en la pared abdominal, 1 en el miocardio, 1 en la pared torácica, 3 en la columna espinal y 1 en el muslo. La hospitalización duró entre 9 y 88 días, con un promedio de 27. Sólo murieron dos pacientes en el período postoperatorio, uno con hidatidosis cardíaco y otro con hidatidosis de la columna. Tres pacientes con múltiples quistes en el peritoneo y uno con quistes en el muslo presentaron recurrencia de la enfermedad y fueron reoperados en forma exitosa.

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Invited Commentary

Mongi Zlitni, M.D.

Service d'Orthopédie Traumatologie, EPS Charles Nicolli, Tunis, Tunisia

The authors present an interesting series of patients with uncommon locations for echinococcasis. Their diagnostic and therapeutic approach is excellent, and their therapeutic results are honorable given the malignant nature of hydatic lesions.

Hydatid disease is characterized by its long clinical latency, allowing only late diagnosis and permitting the disease to become extensive. The diagnosis is not always easy, as many of the manifestations of hydatid disease are similar to those of other diseases—infectious, tumoral, dystrophic—particularly those at osseous sites.

Modern methods, hydatid immunology more than the outdated intradermal Casoni's test, radiographs in certain cases, CT scans and magnetic resonance imaging (MRI) enable us to determine the diagnosis, which is often established only during the surgical intervention.

The treatment must be surgical, as medical treatment has been shown to have limited efficiency, particularly concerning osseous sites. If the treatment is likely to generate recovery from the lesions in the soft parts or in organs such as the kidney, peritoneum, or others, it faces rupture of hydatid cysts in the osseous tissues, rendering surgical removal sometimes impossible. In the spine, what must be undertaken, rather than curettage, is whole vertebrectomy, whenever possible, with metallic instrumentation and bone grafting.

In conclusion, this article reports a good series. It was conducted by authors living in a country bordering the Mediterranean Sea around which hydatic disease constitutes a major concern for the practitioner.