Diaphragmatic Mesothelial Cysts in Children: Radiologic Findings and Percutaneous Ethanol Sclerotherapy

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OBJECTIVE. We describe CT, MR, and sonography findings of diaphragmatic mesothelial cysts and the results of percutaneous treatment with ethanol. All cysts were bilobulate and showed extrahepatic location between the right liver lobe and diaphragm.

CONCLUSION. Radiologic findings are helpful in diagnosing diaphragmatic mesothelial cysts, which should be managed conservatively. Percutaneous ethanol sclerotherapy should be the first choice of treatment if necessary.

Diaphragmatic mesothelial cysts are rare congenital lesions that are lined with mesothelial cells. A limited number of patients, mostly adults, with diaphragmatic mesothelial cysts that were treated by surgery has been reported [1–5]. The diagnosis of diaphragmatic mesothelial cysts might be problematic because of their rarity and their anatomic location.

In this retrospective study, we describe the radiologic findings, including sonography, CT, MRI, and long-term follow-up of children who had diaphragmatic mesothelial cysts with and without percutaneous treatment.

Materials and Methods

Records of 11 children (six girls, five boys) with 11 diaphragmatic mesothelial cysts were retrospectively reviewed, and information about age, lesion size, radiologic findings, treatment, and follow-up was extracted. Patients included in the study were diagnosed between January 1998 and February 2004 at our clinic. The mean patient age was 8 years (range, 3.5–12 years). All cysts were detected incidentally on sonographic examination in patients with nonspecific abdominal pain. Six children underwent CT, and two children underwent MRI. Five children were treated with diagnostic aspiration and ethanol sclerotherapy, and the remaining six children were managed with follow-up sonography.

Informed consent was obtained before diagnostic aspiration and sclerotherapy from all patients. The procedure was performed on an inpatient basis with the guidance of sonography and fluoroscopy and with the patient under IV sedation. The patients were placed in the lateral decubital position, and the location of the diaphragmatic mesothelial cyst was confirmed using sonography in two planes. All patients were monitored during the procedures. After the puncture site was selected and infiltrated with prilocaine HCl, the initial puncture of the cyst was performed using an 18-gauge Seldinger needle with the free-hand technique under sonographic guidance. When the needle reached the cavity, a small amount of clear yellowish fluid was aspirated and sent for bacteriologic, cytologic, and biochemical examinations. The possibility of a hydatid cyst was also investigated. Then a cystogram was obtained to evaluate the shape of the cyst and the presence of extravasation by using Telebrix ([ioxithalamate] 350 mg I/mL, Guerbet). After the absence of extravasation or any fistulous communication was verified, all the cyst contents were aspirated; and 95% ethanol, which was 40% of the initial cyst volume, was injected into the cavity via the needle. Ethanol was left in the cavity for 10 min. Then all ethanol was aspirated and the needle was withdrawn. Patients were called for periodic sonographic examinations 1, 3, 6, and 12 months after the procedure, and once every year thereafter. Three dimensions were measured, and the volume of the cyst was calculated at every visit.

Results

Results were obtained by comparing the cyst volumes calculated at the first and last visits. The overall mean follow-up period was 22 months (range, 3–66 months) (Table 1).

Radiologic findings showed that all the cysts were located between the posterolateral aspect of the right liver lobe and the diaphragm (Figs. 1–3). On sonography in all children, bilobulate cystic structures (Fig. 1) with thin walls and thin hyperechoic lines in

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the wall were identified. High-frequency sonography showed the extrahepatic locations of the cysts and the appearance of the "extrahepatic" sign, which is similar to the extrapleural sign (Fig. 1B). In six children, CT showed homogeneous, nonenhancing, well-defined cysts of water density (Fig. 2). MRI of two children showed thin-walled cystic structures attached to the diaphragm wall and appearing hypointense on T1-weighted and hyperintense on T2-weighted images (Fig. 3). All cysts were bilobulate, calcification of the cyst wall was observed in one cyst, and in one cyst internal echoes were detected with a high-frequency transducer.

For the five cysts treated percutaneously, the mean volume of aspirate was 19 mL (range, 8–36 mL). Average volume reduction after sclerotherapy was 99% during the follow-up period, which ranged from 3 to 66 months (mean, 30.4 months). Cysts disappeared completely in 80% (4/5) of children in this group 1 year after the procedure. In the fifth patient, the rate of volume reduction was 97% 3 months after the procedure. Pain resolved in one patient and decreased in another patient in this group. The cytologic examination was consistent with a mesothelial cyst in one patient who showed mesothelial cells; however, in the remaining cysts, histiocytes and macrophages were observed. Microbiologic examinations were unremarkable, and no scolices of hydatid cysts were detected.

Six other children with similar radiologic findings were managed conservatively with sonographic follow-up. The mean calculated volume of the cysts in this group was 9 mL (range, 2–23 mL). In this patient group managed conservatively, the average volume reduction was 38.3% during the follow-up period (range, 3–48 months; mean, 15 months). In two cysts of two children, no change in size was observed; however, four other cysts showed a decrease in size, with volume reduction rates of 12%, 44%, 80%, and 97%. No increase in size was detected in any of the cysts.

Discussion

Diaphragmatic mesothelial cysts arise from coelomic remnants. Mesothelial cysts may also be detected in organs such as the spleen, adrenal gland, ovary, falciform ligament, vaginal process of the testicle, and mesentery [5]. The bilobulate appearance of diaphragmatic mesothelial cysts may be explained by the complex embryologic development of the diaphragm [6, 7]. Mayer et al. [8] reported a case of a benign two-cavity cystic lesion that had a radiologic appearance similar to that of a diaphragmatic mesothelial cyst and was treated surgically. After histologic examination, the lesion was thought to be either a cyst of the mesonephros or a mesothelial cyst of the diaphragm [8]. In that case report, several explanations were described for the configuration of the cyst, such as embryologic development of the diaphragm, smaller channels through the diaphragm for nerves and blood vessels, the presence of small embryologic defects in the diaphragm, and the cyst primarily originating from the diaphragm [8]. However, for the cysts in our study, the last possibility seems to be most likely because of their anatomic location.

Because of its rarity and the difficulty of identifying its exact anatomic location, a diaphragmatic mesothelial cyst may be misdiagnosed as an intrahepatic simple cyst, a hydatid cyst, or another cystic lesion adjacent to the diaphragm, such as bronchogenic cyst, teratoma, or hydatid cyst.

Estaun et al. [5] reported the largest patient group in the literature, which included four children who underwent surgical treatment of diaphragmatic mesothelial cysts and five children with five cystic lesions who were managed with radiologic follow-up. The same location and radiologic findings of these five cystic lesions led those authors to conclude that the finding of a thin-walled cystic lesion between the liver and the right thoracic wall in the posterolateral aspect of the right costophrenic angle was suggestive of a diaphragmatic mesothelial cyst [5]. Several cases have also been reported of diaphragmatic mesothelial cysts with radiologic findings similar to those reported in the study by Estaun et al. [1, 2, 9]. In our study, 11 cystic lesions could be diagnosed as diaphragmatic mesothelial cysts with the same radiologic findings and cytologic results after excluding other, more common causes. Additional radiologic findings were bilobulation in all cysts and calcification in one cyst. An extrahepatic location could be identified with a high-frequency transducer in most cases; however, coronal MR images might rarely be necessary. To our knowledge, these radiologic findings and percutaneous treatment of diaphragmatic mesothelial cysts with ethanol sclerotherapy have not been reported previously in the English-language literature.

Aspiration was considered diagnostic of diaphragmatic mesothelial cyst, and the decision to perform ethanol sclerotherapy was made on the basis of the imaging findings and the nature of the cyst contents. Ethanol was chosen as the sclerosant agent because it has been commonly used for sclerosing hepatic, renal, and splenic hydatid cysts and lymphoceles safely and successfully [10–14]. In our study, we obtained a high rate of success with ethanol sclerotherapy of diaphragmatic mesothelial cysts without any side effects related to ethanol. Four (80%) of five diaphragmatic mesothelial cysts disappeared completely 1 year after the procedure, and in the other patient the rate of volume reduction was 97% 3 months after the procedure. In this latter pa-

TABLE 1: Demographic Data from 11 Patients with Diaphragmatic Mesothelial Cysts

Patient No.	Age (yr) / Sex	Initial Cyst Volume (mL)	Ethanol Sclerotherapy	Follow-Up (months)	Cyst Volume After Intervention or Radiologic Follow-Up (mL)	Volume Reduction (%)
1	3.5 / F	36	Yes	66	0	100
2	7 / M	8	Yes	44	0	100
3	12 / F	25	Yes	26	0	100
4	12 / M	9	Yes	13	0	100
5	10 / M	18	Yes	3	0.6	97
6	10 / M	5	No	48	1	80
7	7 / F	2	No	18	2	0
8	6 / F	3	No	12	0.1	97
9	6 / M	5	No	5	5	0
10	6 / F	23	No	5	20	12
11	8 / F	16	No	3	9	44

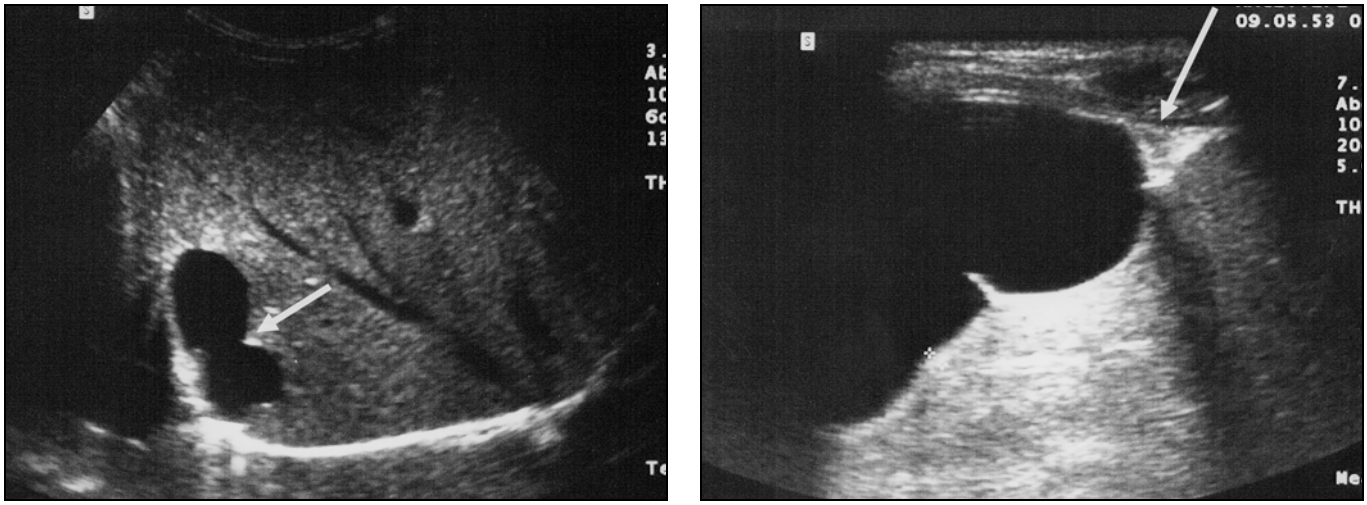


Fig. 1—Sonography findings of diaphragmatic mesothelial cyst in 7-year-old boy.
A, Sonogram obtained with 3.5-MHz transducer shows bilobulate cystic lesion (*arrow*) with thin hyperechoic line in wall.
B, With high-frequency transducer, extrahepatic location (*arrow*) can easily be seen.

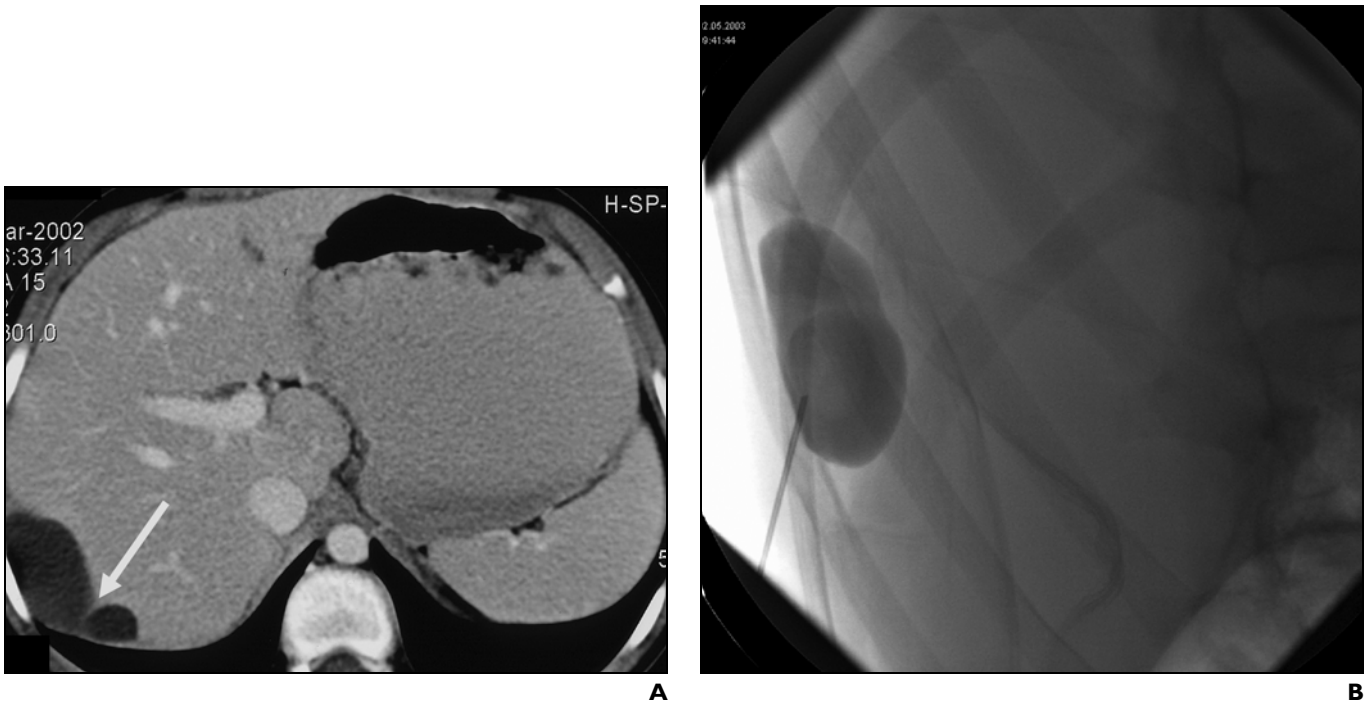
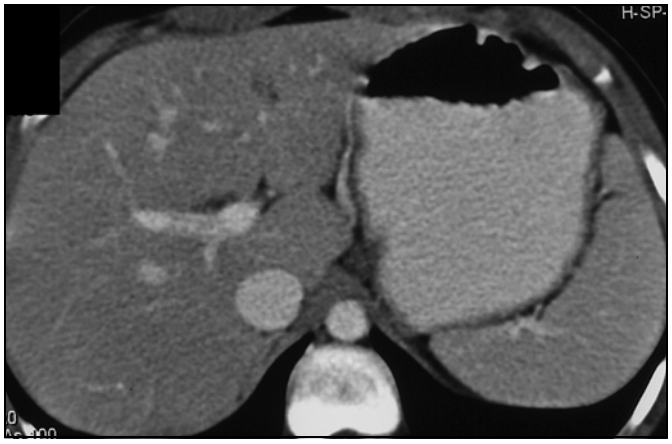


Fig. 2—CT findings of diaphragmatic mesothelial cyst in 12-year-old girl.
A, Pretreatment axial CT image shows bilobulate cystic lesion (*arrow*) adjacent to posterolateral aspect of liver.
B, Cystogram obtained during ethanol sclerotherapy confirms bilobular nature of lesion.
(Fig. 2 continues on next page)

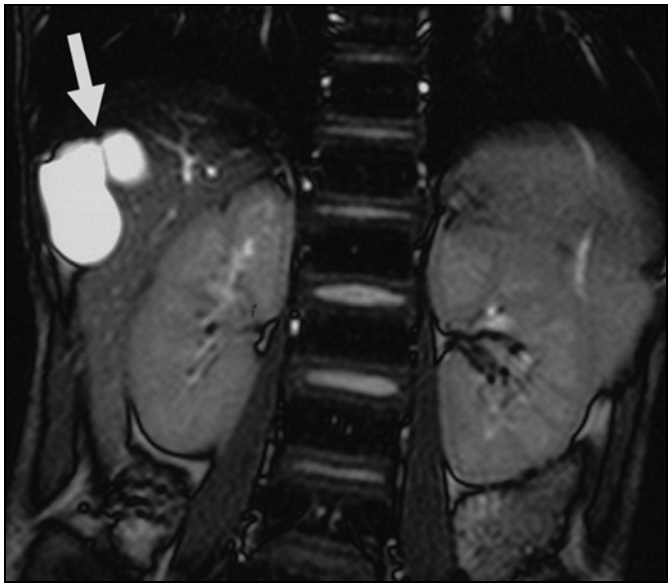
tient, we believe that complete resolution of the cyst will occur by the end of the first year. Although pain resolved in one patient and decreased in another after percutaneous treatment, we are not convinced of a direct relation between pain relief and percutaneous treatment.

Although diaphragmatic mesothelial cysts can be treated surgically and percutaneously, these cysts in children can be managed appropriately with radiologic follow-up using periodic sonographic examinations. Estaun et al. [5] followed up five cystic lesions consistent with diaphragmatic mesothelial cysts for 1–6 years and observed no change in three of



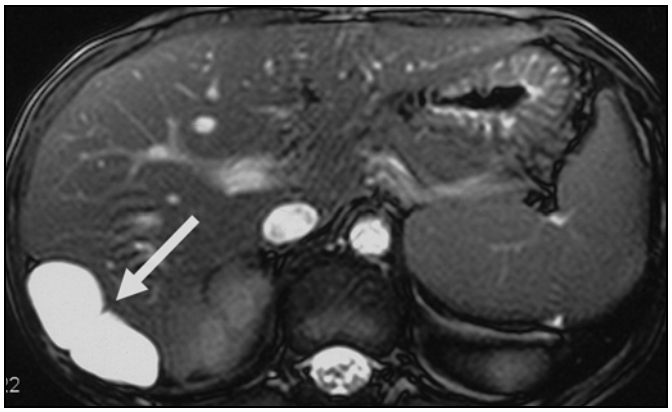
C

Fig. 2 (continued)—CT findings of diaphragmatic mesothelial cyst in 12-year-old girl. **C,** After 9 months of percutaneous treatment, CT image at same level as **A** shows complete resolution of diaphragmatic mesothelial cyst.



A

Fig. 3—MRI findings of diaphragmatic mesothelial cyst in 8-year-old girl. **A** and **B,** T2-weighted coronal (**A**) and axial (**B**) images show bilobulate hyperintense lesion (*arrow*) between diaphragm and right lobe of liver.



B

them, whereas the other two resolved completely, possibly because of a breakdown or collapse of the cyst. We have also seen no change in two of the six cysts and a significant decrease in size in the remaining four. Because of these results, we recommend following up these patients with periodic sonographic examinations. However, if the cyst is symptomatic (pain, secondary infection), percutaneous ethanol sclerotherapy should be preferred instead of surgery because of its safety and high rate of success.

Limitations of our study are its retrospective nature and the lack of pathologic proof in all cases. However, our study is the largest series in the literature and can be a guide in the management of this rare benign disorder.

In summary, we think that in cystic lesions located at the posterolateral aspect of the right lobe of the liver, specific radiologic findings such as bilobulation of the cysts and an extracapsular location of the liver (extrahepatic sign), as described in this article, are pathognomonic findings of diaphragmatic mesothelial cysts. We believe that lesions with this pattern should be managed conservatively with periodic sonographic follow-up; if treatment is needed, percutaneous treatment should be the first choice of technique.

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