

A Tale of Two Cysts: Disparate Outcomes of Two Hepatic Cysts

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Abstract

Simple hepatic cysts occur in less than 5% of the population. As the technology we use for imaging advances, so does the frequency with which we identify them. Simple cysts, as presented here, do not generally require aspiration. The majority of these cysts do not require treatment, however, those larger than 4 cm are recommended for surveillance. We present two cases of simple cysts that were followed for 15 years and managed differently. The experience we acquired from these cases illustrates challenges in understanding and management and an implication that, perhaps, conservative surveillance might be best.

Case 1

A 76-year-old male with chronic hepatitis B was noted on surveillance ultrasonography to have an asymptomatic right lobe hepatic cyst measuring 7.3 × 7.3 × 6.3 cm in 1998. While undergoing anti-HBV therapy, he underwent surveillance imaging annually. In 2001, the cyst had increased to 9.1 × 9.0 × 8.5 cm. In 2005, the cyst was noted to increase to 10 × 9.5 × 9.0 cm. At that time, mild intrahepatic ductal dilatation was noted, but the patient remained asymptomatic with normal liver function. Throughout this surveillance period, his chronic hepatitis B was well controlled with antiviral therapy. After discussion of treatment options, the decision was made to manage the cyst conservatively with continued surveillance [1]. Over the next five years, annual MRI or ultrasound showed no change in size. The patient remained asymptomatic. In 2010 (12 years later), the cyst decreased in size to 5.5 × 6.4 × 3.4 cm. In 2012, the cyst decreased to 4.5 cm and, by 2013, it measured only 2.4 cm. Resolution of the intrahepatic ductal dilatation was also noted. In summary, over the course of 15 years of surveillance, this patient's hepatic cyst increased to a maximum size of 10 cm, without ever causing symptoms or lab abnormalities, only transient ductal dilatation and underwent spontaneous regression without intervention (Figure 1).

Case 2

A 58-year-old woman developed non-Hodgkin's lymphoma of the left submandibular node in 1998. On surveillance imaging of her known lymphoma, the patient was found to have a 4.5 cm right lobe hepatic cyst. For her lymphoma, she ultimately underwent resection, radiotherapy and chemotherapy. In follow-up abdominal imaging thirteen years later in 2011, her hepatic cyst was noted to have increased to 9.4 × 8.5 cm. The patient was completely asymptomatic, but concerned given its doubled size and sought further consultation. She elected to undergo cyst aspiration with 350 ml of simple fluid followed by alcohol sclerotherapy. Three months later, follow-up imaging showed a decrease to 7 × 6.8 × 5.7 cm. One year later in 2012,

the cyst increased to 7.3 × 6.6 × 6.0 cm. In 2013, imaging showed the cyst increased to 10.4 × 9.1 cm, beyond its size at initial diagnosis. Due to symptoms of abdominal fullness, the patient again underwent aspiration of 520 ml of simple fluid with sclerotherapy. One year later, in 2014, despite now being asymptomatic, imaging showed the cyst returned to a size of 9.6 × 9.4 cm. Since she remained asymptomatic, and had now failed two trials of aspiration with sclerotherapy, she elected to continue only with conservative surveillance monitoring. In summary, over the course of 15 years of surveillance, this patient's hepatic cyst with mild transient symptoms underwent aspirations, but without lasting benefit and a cyst that remained at its original size (Figure 2).

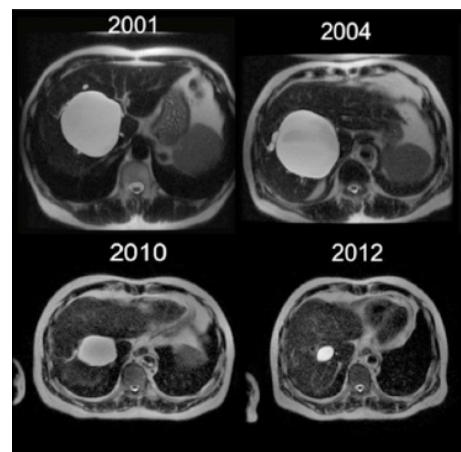


Figure 1: Images showing surveillance MRI scans from 2001 through 2012 of case 1, with spontaneous regression of the hepatic cyst without intervention. At its largest the cyst was 10 × 9.5 × 9.0 cm, but regressed to 2.4 cm.

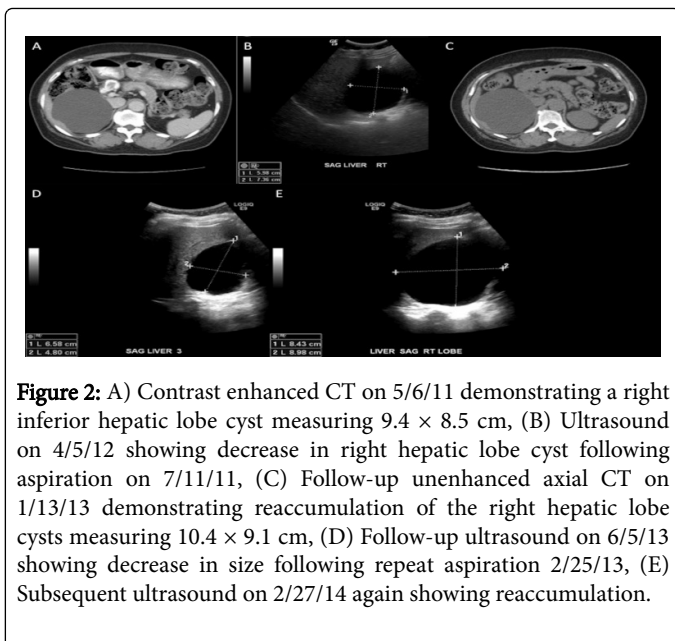


Figure 2: A) Contrast enhanced CT on 5/6/11 demonstrating a right inferior hepatic lobe cyst measuring 9.4 × 8.5 cm, (B) Ultrasound on 4/5/12 showing decrease in right hepatic lobe cyst following aspiration on 7/11/11, (C) Follow-up unenhanced axial CT on 1/13/13 demonstrating reaccumulation of the right hepatic lobe cysts measuring 10.4 × 9.1 cm, (D) Follow-up ultrasound on 6/5/13 showing decrease in size following repeat aspiration 2/25/13, (E) Subsequent ultrasound on 2/27/14 again showing reaccumulation.

Discussion

While these two cases by clinical, radiologic and chemical markers were not concerning for infection, that distinction is not always easy to make. The differential for intrahepatic cysts includes infection and must be excluded. Clinically, there are clues in deciphering an infectious from a non-infectious cyst. After infection, the time to symptomatic presentation is 2-5 months and generally starts as right upper quadrant pain, though sometimes referred, with fever [2]. Diarrhoea can occur in conjunction, but in one report was only seen in one-third of patients [3]. Various other symptoms have been reported, such as night sweats, jaundice, fatigue, weight loss, poor appetite, cough and hiccoughs. Exam typically reveals an enlarged liver and tenderness to palpation in the right upper quadrant. Rupture can occur in up to 7% of cases into the peritoneum, causing peritonitis, or even more commonly into the chest [4]. Amoebic liver abscesses can radiographically appear similar to simple cysts as round, well-defined hypoechoic masses [5]. On CT scan, however, they can have a characteristically peripherally enhancing rim and, on MRI, demonstrate different characteristic signal intensities-low on T1-weighted and high on T2-weighted images. They occur most commonly in the right lobe and a majority are solitary subcapsular lesions [6]. Pyogenic liver abscesses are also difficult to distinguish from cysts on ultrasound. On CT, however, features of stranding or loculated subcollections can help identify a pyogenic etiology. In cases of pyogenic liver abscess, drainage and culture should be obtained to guide therapy, as should blood cultures since they are positive in almost 50% of cases [7]. On routine labs, a leukocytosis without eosinophilia is common in *Entamoeba histolytica* whereas in echinococcus a leukopenia with eosinophilia is seen. Liver function tests in all abscess etiologies reveal an elevated alkaline phosphatase and, sometimes, elevated aminotransferases. Serologic testing for *E. histolytica* is useful, since over 90% of infected patients have detectable antibodies at presentation, though they may be negative during the first week of infection [8]. Antibodies are more sensitive than antigen for detection. As with many serologies, it is most useful to exclude disease, but cannot distinguish current and previous infections.

Diagnosis of amoebic abscess does not typically require needle aspiration, unless there is an unclear diagnosis, risk of impending rupture or clinical worsening despite appropriate treatment [9].

While the diagnosis of simple hepatic cyst was not difficult in these patients based on their clinical and radiologic presentation, they did prove to be an interesting challenge in management. These two cases illustrate the difficulty in management with somewhat paradoxical outcomes. In both instances, the patients were monitored over 15 years, which has not been previously reported in the literature. The former case demonstrated spontaneous regression without intervention, while the latter recurred even after two aspirations with alcohol sclerotherapy. The question must be asked, in an asymptomatic or even mildly symptomatic patient, is an intervention actually changing the natural course of the cyst or merely addressing the anxiety of the patient and physician?

Despite different comorbid conditions in these patients, hepatitis B and non-Hodgkin's lymphoma, there is no association with the formation of simple cysts and either of these diseases. As mentioned above, the vast majority of simple hepatic cysts can be managed without any intervention. Large cysts (>4 cm) can be monitored with serial imaging, which is typically sufficient after two years provided that the cyst remains unchanged in size [10]. It is when they become symptomatic, most commonly causing abdominal discomfort, distention, early satiety, nausea, or vomiting, that intervention becomes a consideration.

Traditionally, the management of these simple cysts, when necessary, involves percutaneous ultrasound, CT-guided drainage or surgical unroofing. Aspiration alone has vastly fallen out of favour, due to high recurrence rates, approaching 78-100% [11,12]. Consequently, simple aspiration has been replaced with combination percutaneous drainage and alcohol sclerotherapy, as in our second patient. In this method, the cyst is aspirated, and then injected with a sclerosing agent, such as ethanol. The goal of sclerosant is to disable the cyst lining from further secretion and subsequent enlargement [13]. In a number of case series and reports, alcohol sclerotherapy has been reported as safe and effective [14,15]. While success rates with alcohol sclerotherapy typically approach 100%, the recurrence rates approach 20% after as little as 4 months, albeit markedly smaller, ranging from 30-50% of the original cyst size [16]. While other options for intervention exist, such as conventional or laparoscopic deroofing, even though the recurrence rates decrease for each procedure, the morbidity increases, so these options are not without risk [17].

Literature that pertains to our patient with spontaneous regression is especially sparse. This is not to say spontaneous regression is unseen, rather potentially underreported, as anecdotal experience of an expert physician at our institution notes frequent incidental findings of hepatic cysts in the range of 3-4 cm that remain unchanged for years. Arai et al. reported a case of a 55-year-old woman with a 7.7 cm simple hepatic cyst that underwent spontaneous regression to 1 cm with no treatment [18]. The only other case report of spontaneous hepatic cyst regression was a congenital cyst in a 6-week-old infant [19]. The remaining literature that discusses the disappearance of cysts, typically hydatid cysts, attributes resolution to either fistula formation into the biliary or enteral tracts or intraperitoneal rupture [20,21]. Our patient had no evidence of rupture or fistula. One possibility might be extrapolated from literature discussing the rare spontaneous regression of HCC. There is evidence to suggest that a change in the dynamic of blood flow to the tumor, or possibly cyst, limits further growth and

regression occurs as sequelae. Though the literature on regression is rare, maybe the incidence is not as rare as we think.

In summary, simple hepatic cysts are a relatively benign hepatic malformation; however, they pose a challenging clinical scenario. Given their disparate presentations and clinical course, it is difficult to ascertain a treatment algorithm without a better understanding of their pathogenesis. Our case histories pose a unique juxtaposition-one favoring surveillance, while another faced the risk of interventions without ultimate benefit. Without signs of worsening symptoms, infection, rupture, or malignancy, these cases highlight not only the safety of a conservative approach, but possibly a more favourable outcome. In addition, while literature on various intervention recurrence rates is easier to find, we suggest a closer look at the rate of stability or regression of large cysts to better understand who would require a more aggressive approach. A personalized approach remains the best compromise of risk and benefit, but we recommend these cysts still be managed cautiously and conservatively when able.

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