Nonparasitic liver cysts: different treatment strategies

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Objective
The aim of this work was to study the clinicopathological data of nonparasitic liver cysts (NPLCs), its different management modalities, and outcome.

Patients and methods
This retrospective study included patients who were diagnosed as having NPLCs from January 2000 to the start of 2016. The clinicopathological data, surgical and nonsurgical treatment, and outcomes of these patients were studied.

Results
NPLC was present in 118 patients. There were 78 (66.1%) female patients and the median age was 48 years. The majority of cases (95; 80.5%) were of simple liver cysts and its management was conservative treatment with follow-up (51 patients; 53.7%), percutaneous aspiration, puncture aspiration injection and reaspiration or pig-tail catheter drainage (26 patients; 27.4%), and surgical treatment (18 patients; 18.9%) with either laparoscopic deroofing (12 patients) or open surgery (six patients). Six (5.1%) patients with intrahepatic biloma underwent percutaneous aspiration or pig-tail drainage. Five (4.2%) patients had cystadenoma and underwent resection or pericystectomy. Five (4.2%) patients had post-traumatic hematoma and underwent conservative treatment. Three (2.5%) patients had polycystic liver disease; one of them underwent laparoscopic deroofing of large ones and two patients underwent conservative treatment. Two (1.7%) patients had Caroli's disease and were prepared for liver transplantation. Two (1.7%) patients had cysts with biliary atresia and underwent Kasai operation with excision of the cyst.

Conclusion
Most of the NPLCs are simple liver cyst that can be managed conservatively if it is asymptomatic and small, or with percutaneous radiological intervention or laparoscopic deroofing for large symptomatic or recurrent ones. Open or laparoscopic resection or pericystectomy is reserved for cystic neoplasms which is not common.

Keywords:
cystadenoma, liver cyst, nonparasitic liver cysts, puncture aspiration injection and reaspiration, polycystic liver disease, simple liver cyst

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Background

Liver cysts are classified as true or false depending on the presence of an epithelial lining. True cysts include congenital cysts [simple liver cyst (SLC) and polycystic liver disease (PCLD)], parasitic (hydatid) cysts, neoplastic cysts (including cystadenoma, cystadenocarcinoma, cystic sarcoma, squamous cell carcinoma, and metastatic ovarian, pancreatic, colon, renal, and neuroendocrine cancers), and biliary duct-related cysts (Caroli's disease, bile duct duplication, and peribiliary cysts). False cysts may be caused by spontaneous intrahepatic hemorrhage, post-traumatic hematoma, or intrahepatic biloma [1,2].

SLCs are believed to be congenital in origin and arise from aberrant bile ducts. These cells secrete a fluid with water and electrolyte content similar to that of serum [3].

SLCs are small in most patients but can grow to over 30 cm in selected cases. In a small fraction of patients, symptoms such as abdominal pain, early satiety, nausea, and vomiting arise as a result of a mass effect. Physical examination may reveal a palpable abdominal mass or hepatomegaly [4].

The management of most SLC is conservative. If there are symptoms, aspiration–sclerotherapy or surgical (laparoscopic or open) deroofing is the preferred treatment [4].

Cystadenoma is a biliary cyst tumor arising from biliary epithelium [5]. Compared with its malignant counterpart (cystadenocarcinoma), it accounts for less than 5% of all cystic lesions of the liver, but it is dangerous due to its propensity toward local recurrence and

malignant change [6,7]. Cystadenocarcinoma can arise de novo or from a pre-existent cystadenoma, from which it is difficult to differentiate [8]. There is no definite, reliable criterion for differentiating cystadenoma from cystadenocarcinoma, and the correct diagnosis is often made only with histopathology [5].

Caroli's disease is a congenital autosomal recessive malformation characterized by diffuse or segmental cystic dilation of the intrahepatic biliary system. In Caroli's syndrome, periportal congenital fibrosis or multicystic renal diseases are observed in addition to biliary dilations [5,9].

PCLD is arbitrarily defined as the presence of more than 20 liver cysts [10,11]. Liver function, as judged by parameters of liver synthesis, is not affected in PCLD, as functional hepatic tissue remains unaffected [12]. The main objective of therapy is to reduce liver cyst volume to diminish mass effect-related symptoms [13]. Aspiration–sclerotherapy and fenestration are indicated when PCLD consists of large cysts confined to a limited part of the liver. Segmental hepatic resection or even liver transplantation is essential in some cases [4,14].

Biliary atresia is an inflammatory condition of the immature extrahepatic and intrahepatic biliary tract with progressive obliteration of the bile ducts. Cystic biliary atresia is an uncommon form of biliary atresia with an incidence of ~10%. Surgical management (Kasai portoenterostomy) aiming at restoring bile flow to the bowel should be performed ideally before 40 days following birth [15].

The aim of this work was to study the clinicopathological data of nonparasitic liver cyst (NPLC), its different management modalities, and outcome.

Patients and methods

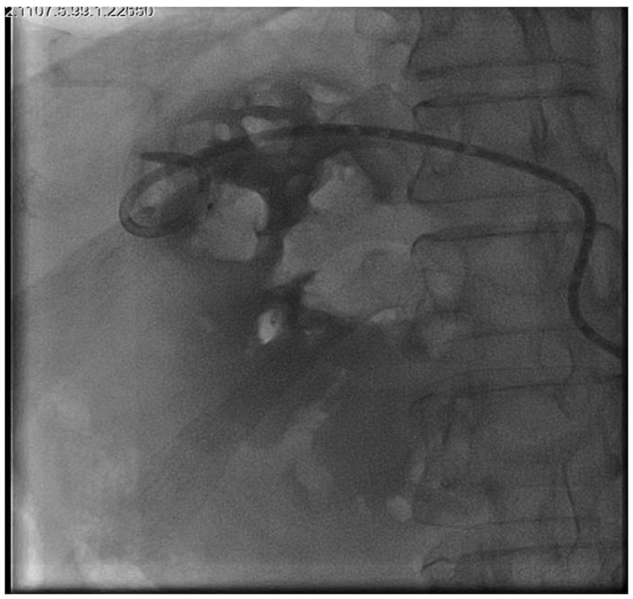
This retrospective study included patients who were diagnosed as having NPLCs and presented to our institute from January 2000 to the start of 2016. All cases of parasitic (hydatid cysts) or infectious (pyogenic or amebic) hepatic cystic lesions were excluded from this study.

The clinical data, operative and nonoperative treatment, and outcomes of these patients were studied. The study was approved by our Institutional Ethical Committee. The data collected included age and sex, symptoms and signs, and the presence of any acute complications. To be sure that the NPLC is symptomatic, a lot of investigations were carried out for many patients, including upper endoscopy and colonoscopy. Conservative management of small (<5 cm in diameter) or asymptomatic NPLC was carried out with follow-up clinically and with abdominal ultrasound.

The patients treated with interventional radiology were treated at the start of the period with aspiration. There was a high rate of recurrence. Therefore, most of the patients were treated using the puncture aspiration injection and reaspiration (PAIR) technique, which was described originally for hydatid cystic disease of the liver [16]. This was preceded by injection of contrast material inside the cystic cavity to ensure no communication with the biliary tree before injection of the sclerosant material (Fig. 1). It is the same as aspiration and injection of sclerosant material, which was already described for NPLC [17]. PAIR was performed under local anesthesia and ultrasound (US) guidance. An overall 25% of the volume was replaced with 95% alcohol and then completely aspirated after 20 min. Pig-tail catheter drainage alone as a line of treatment was performed for only few patients.

Laparoscopic deroofing of NPLC was carried out, with special emphasis on hemostasis and removal of a part of the normal hepatic tissues adjacent to the dome of the cyst to decrease the recurrence rate [18,19]. Open deroofing, pericystectomy, and hepatic resection were performed in the classic way.

Figure 1



No biliary communication after injection of contrast inside simple liver cyst.

Table 1 Patient characteristics according to the types of nonparasitic liver cysts

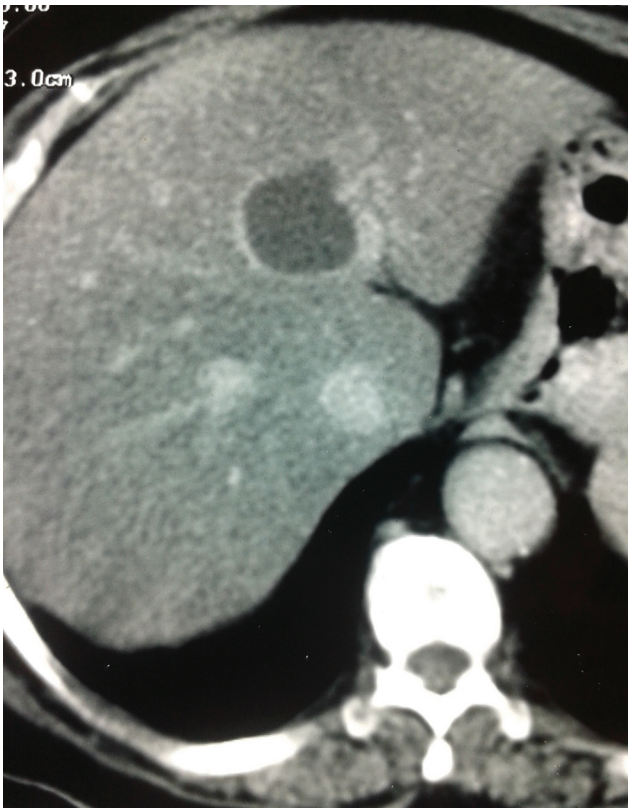
Types of NPLC	*N* (%)	Male [*n* (%)]	Female [*n* (%)]	*P* value
SLC	95 (80.5)	25 (26.3)	70 (73.7)	0.004
Cystadenoma	5 (4.2)	1 (20.0)	4 (80.0)	
PCLD	3 (2.5)	1 (33.3)	2 (66.7)	
Caroli's disease	2 (1.7)	1 (50.0)	1 (50.0)	
Cystic biliary atresia	2 (1.7)	2 (100.0)	0 (0.0)	
Intrahepatic biloma	6 (5.1)	5 (83.3)	1 (16.7)	
Post-traumatic hematoma	5 (4.2)	5 (100.0)	0 (0.0)	
Total	118 (100)	40 (33.9)	78 (66.1)	

There is a highly statistical significance ($P<0.01$) between male and female patients with SLC than the total number of other patients with NPLC. NPLC, nonparasitic liver cyst; PCLD, polycystic liver disease; SLC, simple liver cyst.

Table 2 Clinical presentation of patients with simple liver cyst

Clinical presentations	Number of patients [*N* (%)]
Asymptomatic	35 (36.8)
Abdominal pain	29 (30.5)
Hepatomegaly	16 (16.8)
Abdominal mass	11 (11.5)
Jaundice	8 (8.4)
Chest pain	6 (6.3)
Early satiety	5 (5.2)
Dyspnea	3 (3.1)
Nausea or vomiting	3 (3.1)

Figure 2



Computed tomography scan of recurrent simple liver cyst after aspiration.

Outcome parameters in the form of early and late complications and any mortality or recurrence were reviewed. The follow-up was carried out with US every 3 or 6 months and with computed tomography scan every 6 or 12 months according to the diagnosis.

Statistical analysis

Data were statistically analyzed using statistical package for the social sciences (SPSS) program (version 21 for Windows; SPSS Inc., Chicago, Illinois, USA). A *P* value of less than 0.05 was considered statistically significant. Data were shown as frequency and percent. The χ^2-test was used for all these qualitative variable analyses and Student's *t*-test was used for quantitative data analysis.

Results

NPLC was present in 118 patients. There were 78 (66.1%) female patients and the mean age was 48 years (Table 1). Laboratory investigations, serology, and tumor markers were normal in most of the patients. Cases of jaundice showed variable increase in serum bilirubin and other cholestatic parameters. Only two cases of cystadenoma showed mild elevation of CA 19-9.

In our patients who were treated surgically or with interventional radiology, the hepatic cysts measured 5–25 cm, and all were symptomatic.

The majority of cases (95; 80.5%) were of SLC as shown in Table 1, which also shows the various types of NPLC and the number, percent, and sex of each group of patients. The various clinical features of SLC are shown in Table 2, and abdominal pain was the most frequent presentation. Figure 2 shows the computed tomography scan picture of recurrent SLC after aspiration. The management of SLC is shown in Table 3 in the form of conservative treatment with follow-up (51 patients; 53.7%), percutaneous aspiration, PAIR, or pig-tail catheter drainage (26 patients; 27.4%), and surgical treatment (18 patients; 18.9%) either with laparoscopic deroofing (12 patients), or open surgery (six patients). The SLC

Table 3 Different management modalities of simple liver cyst and rate of recurrence

Procedures	Number of patients [*N* (%)]	Recurrence [*N* (%)]	*P* value
Radiological	26 (27.4)	9 (34.6)	0.03
PAIR	16 (16.8)	3 (18.7)	
Aspiration	6 (6.3)	4 (66.6)	
Pig-tail drainage	4 (4.2)	2 (50)	
Surgical	18 (18.9)	1 (5.5)	
Lap deroofing	12 (12.6)	1 (8.3)	
Open deroofing	4 (4.2)	0 (0)	
Liver resection	2 (2.1)	0 (0)	
Conservative	51 (53.7)	–	
Total	95	10/44 (22.7)	

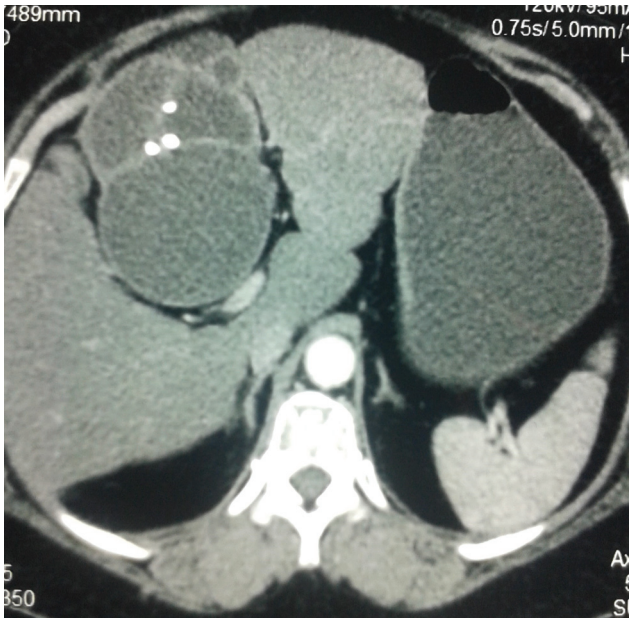
There is a statistical significance ($P<0.05$) for the recurrence between the patients treated surgically and radiologically. PAIR, puncture aspiration injection and reaspiration.

treated conservatively were small and asymptomatic. They did not change in size with a follow-up period of at least 3 months. Many patients were lost to follow-up. We did not encounter any case of complications in the files of this group of patients. Rate of recurrence was significantly higher in patients treated with interventional radiology, especially those with aspiration only (Table 3). One patient showed recurrence after surgical management and was treated conservatively. Nine patients showed recurrence after radiological management. These patients were also treated conservatively as symptoms disappeared or became well tolerated. No recurrence was observed for other types of cysts.

Six (5.1%) patients with intrahepatic biloma underwent percutaneous aspiration or pig-tail drainage. Five (4.2%) patients had cystadenoma, of whom three underwent resection and two underwent pericystectomy (Figs 3–6). Five (4.2%) patients had post-traumatic hematoma and underwent conservative treatment (Fig. 7). Three (2.5%) patients had PCLD; one of them underwent laparoscopic deroofing of large ones, and two patients underwent conservative treatment. Two (1.7%) patients had Caroli's disease and were prepared for liver transplantation. Two (1.7%) patients had cysts with biliary atresia and underwent Kasai operation with excision of the cyst (Fig. 8).

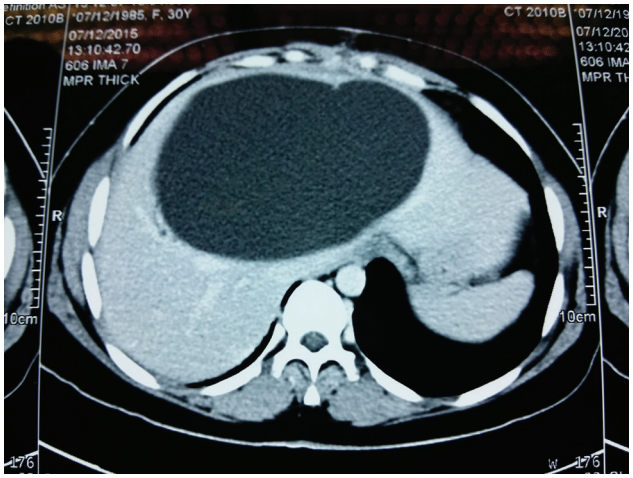
Table 4 shows the difference in morbidity rates between surgical and radiological management of SLCs. There was higher morbidity in patients treated surgically and no mortality in both groups. There was one female patient with cystadenoma who developed chest infection and wound infection after hepatic resection.

Figure 3



Computed tomography scan of hepatic cystadenoma.

Figure 4

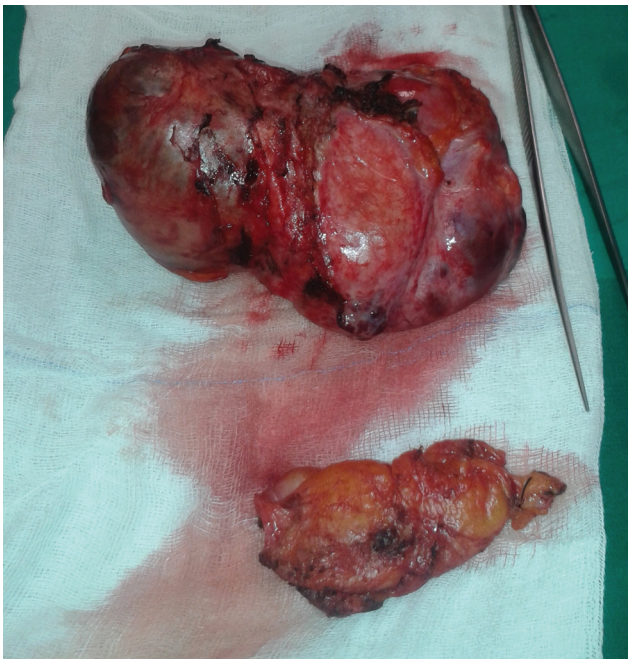


Computed tomography scan of hepatic cystadenoma.

Discussion

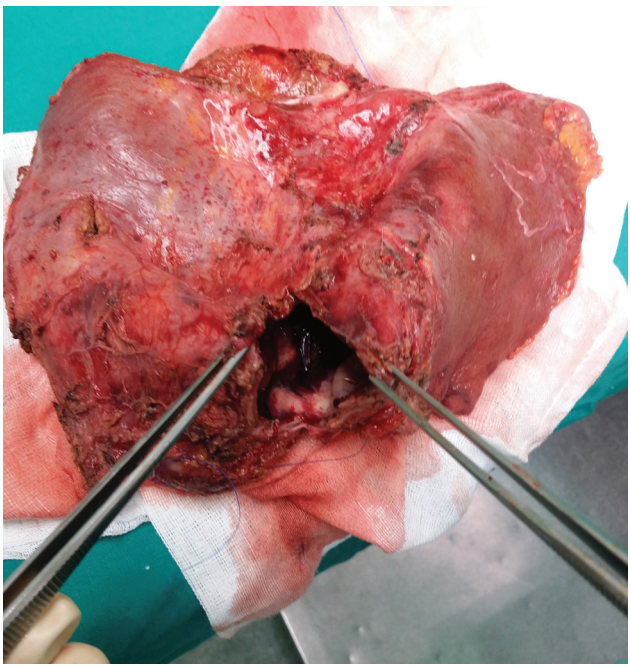
Previous studies, based on autopsy and surgical series, estimated a very low prevalence of simple NPLCs (0.14–0.17%) [20]. More recently, among patients referred for abdominal ultrasonography, the prevalence of SLC has been reported to be 2.5–4.65%. Liver cysts are being recognized increasingly as the routine use of imaging studies becomes more widespread. Hepatic cysts may be more common in women and in patients older than 40 years [21,22]. Symptoms, although quite rare, may be related to the space-occupying effect of large cysts. Symptoms may include abdominal discomfort, chronic right upper quadrant or epigastric abdominal pain, early satiety, dyspnea, increasing abdominal

Figure 5



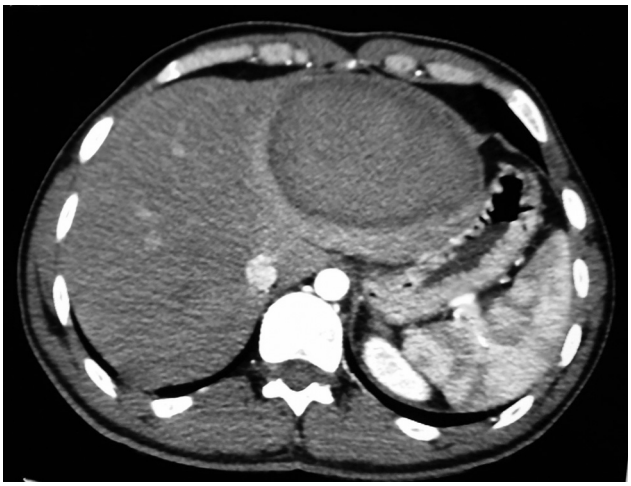
Pericystectomy of hepatic cystadenoma.

Figure 6



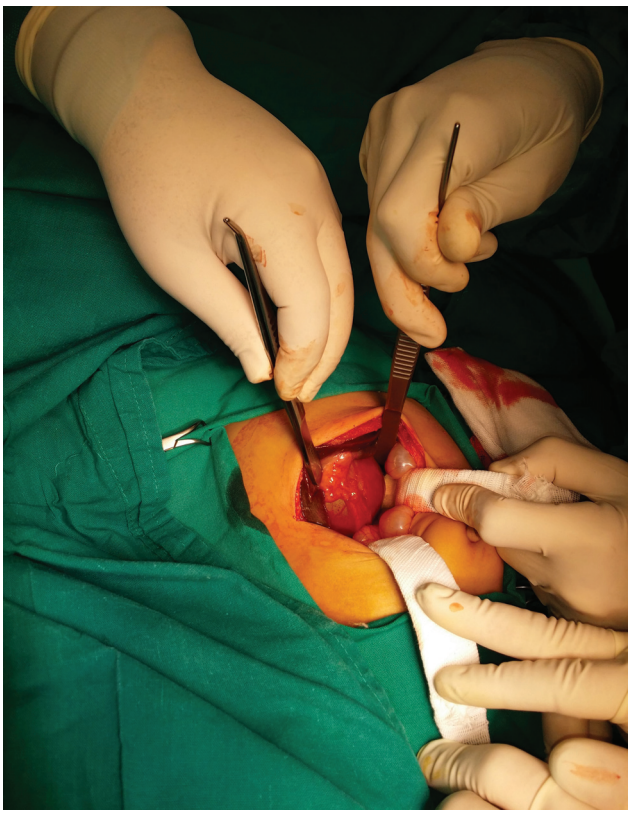
Hepatic resection of hepatic cystadenoma.

Figure 7



Post-traumatic hematoma.

Figure 8



Cystic biliary atresia.

girth, nausea, and vomiting [23]. Sanfelippo *et al.* [20] reported that, among 15 symptomatic patients with solitary NPLCs, abdominal mass was present in 54%, hepatomegaly in 40%, abdominal pain in 33%, and jaundice in 9% of patients [2].

This is in accordance with our results, which showed that NPLCs are more common in women (66%) and the mean age is 48 years. However, false hepatic cysts, such as intrahepatic biloma and post-traumatic hematoma, were more common in men. This is most likely due to the fact that men are more prone to trauma compared with women. In our study, most of the cases were asymptomatic (37%), and abdominal pain, hepatomegaly, and abdominal mass were the most common presentations. It should be stressed that attribution of symptoms to SLC should

Table 4 Morbidity of surgical and radiological management of simple liver cyst

Morbidities	Radiological [*N* (%)]	Surgical [*N* (%)]	*P* value
Bile leak	1 (3.8)	2 (11.1)	0.63
Wound infection	1 (3.8)	2 (11.1)	
Chest infection	0 (0)	3 (16.6)	
Subhepatic collection	1 (3.8)	1 (5.5)	
Total	3/26 (11.5)	8/18 (44.4)	

There is a higher percentage of morbidity among patients treated surgically than radiologically, although not statistically significant ($P>0.05$).

be undertaken with caution after excluding alternative diagnosis [2].

In our study, most of the cases of NPLCs were SLC (80%) and the majority of them were treated conservatively (54%). Hepatic cystadenoma constituted about 4% of cases of NPLC, and mostly occurred in women. False hepatic cysts roughly constituted 10% of our cases of NPLC. The remaining types (PCLD, Caroli's disease, and cystic biliary atresia) were rare and necessitated different treatment modalities.

There were different treatment modalities for SLC in our study with variable results. The interventional radiological management had increased rate of recurrence (35%) compared with the surgical management (6%), which was statistically significant. Aspiration alone had the highest rate of recurrence (67%), whereas open deroofing and liver resection had a zero recurrence rate. High recurrence rates with aspiration alone were shown in most of the previous studies and were near to 100% [24,25].

Several studies demonstrated the efficacy and safety of cyst aspiration followed by injection of sclerosant materials inside the cystic cavity [17,26]. We used in our study the PAIR technique that was already described for hepatic hydatid disease. It has the same principles as aspiration with injection of sclerosant material that already described for SLC. Our recurrence rate with PAIR was only 18%, which was lower compared with aspiration alone or pig-tail catheter drainage. However, this was not statistically significant, most likely because of the small number of patients.

Our study showed good results for laparoscopic deroofing with low recurrence rate (8%). Several recent series have demonstrated good results for laparoscopic deroofing procedures. Widest possible excision of the cystic wall and concomitant argon beam coagulation or electrocoagulation may improve the results. Recurrence has ranged from 0 to 20% with morbidity from 0 to 25% [27–29].

No randomized trials have been conducted comparing sclerotherapy and fenestration for simple cysts. Studies comparing the two treatment methods are small and include heterogeneous groups of patients, but they reported that recurrence rates are higher after sclerotherapy [30].

Our study showed that the complications of surgical management of SLCs were more frequent than that of radiological management. There was no mortality in both groups. In a multicenter survey on 21 patients with SLC treated with aspiration and ethanol injection, there were no complications or deaths, and all symptoms disappeared after treatment [17].

There were five patients with hepatic cystadenoma in our study (4.2% of NPLCs; four female patients, with a mean age of 48 years). All of them were treated by means of open surgery, either pericystectomy or hepatic resection. This is consistent with other series in the literature in which almost all patients with such cystic neoplasms were women in their late 40s. Although some authors have suggested that these cystic neoplasms can only be found in women, other series in the literature suggest that men can account for up to 10% of patients with this pathology [31–34]. We did not encounter cystadenocarcinoma in our study. However, this could be due to the small number of cystic neoplasms. In another study, malignant cystadenocarcinoma was identified in 2/13 (15%) pathological specimens. Both patients presented with small foci of malignant cells within mural nodules that were identified preoperatively [34]. It is widely accepted that these cystic tumors, either benign or malignant, need complete excision [31,32]. Meanwhile, some authors advocate follow-up for risky patients who underwent incomplete excision or laparoscopic deroofing for the benign ones [34].

Conclusion

Most of the NPLCs are SLC that can be managed conservatively if it was asymptomatic and small, or with percutaneous radiological intervention or laparoscopic deroofing for large symptomatic or recurrent ones. However, randomized studies with a large number of cases (if possible) are needed for better understanding the best way of its management. Open or laparoscopic resection or pericystectomy is reserved for cystic neoplasms which is not common.

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Nil.

Conflicts of interest
There are no conflicts of interest.

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