

Nonparasitic Cysts of the Liver: Results and Options of Surgical Treatment

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Abstract. Nonparasitic cysts of the liver (NPHC) are highly variable in respect to appearance and therapeutic approach. The treatment of these cysts varies according to the nature and appearance of the disease. Based on the variable nature of disease and the various therapeutic options, all of which were attempted in our patients, the most suitable mode of treatment for different forms of NPHC are discussed. Ninety-one patients with NPHC who had been treated surgically from 1977 through 1995 were examined retrospectively. Asymptomatic peripheral cysts measuring up to 10 cm do not require further treatment. Computed tomography (CT)guided aspiration (n = 9) should be regarded as a palliative measure. Within a short period, CT-guided aspiration led to recurrence of symptoms in seven of our patients. Standard treatment of NPHC is fenestration with widest possible excision of the cystic wall, which can be performed laparoscopically (n = 10) or by the conventional surgical mode (n = 54). One patient was initially operated by the laparoscopic technique but developed bleeding, which necessitated conversion to the open mode. Three patients underwent synchronous laparoscopic cholecystectomy. Recurrence rates were similar: 11% in the laparoscopically treated group and 13% in the group that underwent conventional open surgery. Conventional surgical treatment was always successful in cases of solitary cysts. However, in cases of multiple cysts measuring more than 5 cm, conventional surgery was followed by recurrence of symptoms in 26% of patients (7/27), who then had to undergo a second operation. Partial resection of the liver (n = 9) was successfully performed in cases of polycystic disease (n = 5) with concomitant enlargement of the organ as well as in cases of large solitary cysts of the left lobe of the liver (n = 4). In patients in whom we found that the cysts communicated with the ductal system (n = 3), we performed a cystojejunostomy to drain the bile. The complication rate was low. In addition to frequent postoperative ascites, which necessitated no further intervention, we observed infectious complications in four patients. Twenty patients (22%) expired during a mean follow-up period of 6.2 years. Interestingly, deaths were frequently associated with malignancy (11/20). After fenestration of multiple cysts measuring > 5 cm, the patients are at high risk for recurrence. Hence partial resection of the liver is an excellent therapeutic alternative in selected patients with polycystic disease and massive enlargement of the organ in whom the disease could not be controlled by simple fenestration. The results of this study show that laparoscopic fenestration should replace the conventional surgical technique as the gold standard in cases of NPHC because the laparoscopic technique is less stressful for the patient and is associated with a rate of success similar to that of the conventional technique.

The development of congenital nonparasitic hepatic and renal cysts is supposedly triggered by chromosome 16. These diseases then presumably represent a single dominantly autosomal genetic entity. The more frequent occurrence of this phenomenon in

women is attributed to the presence of estrogens [1, 2]. Cystic lesions of the liver have been frequently detected during the last decades, both as incidental findings and as a result of specific diagnostic studies. The increase in the number of detected hepatic cystic lesions may also be attributed to improved technical standards and the widespread availability of ultrasonography and computed tomography (CT) units.

Once hepatic cysts are detected, the following diagnoses should be considered: bile duct cysts, parasitic cysts, congenital nonparasitic cysts (NPHCs), or even neoplastic or inflammatory cysts. The morphology of the cysts as depicted by imaging studies and the serologic findings in cases of suspected *Echinococcus* cysts enable the examiner to make appropriate differential diagnoses.

Once the diagnosis of NPHC is established by these techniques, the physician is confronted with a wide spectrum of therapeutic alternatives, ranging from no intervention to percutaneous guided aspiration as well as laparoscopic and conventional surgery. When surgeons decide in favor of conventional surgical treatment, they are again faced with several options: fenestration, excision, partial hepatic resection, or cystojejunostomy. They then must adapt the chosen technique to the respective indication.

The aim of our study was to present and discuss our experience with the various therapeutic modalities in 91 patients. The goal was to find the most effective and safe method, with special emphasis on suitable therapeutic techniques for various forms of NPHC. One aim of the study was to explicate the fact that the inhomogeneity of this disease does not permit general recommendations regarding the surgical approach.

Patients and Methods

A total of 91 patients with NPHC were treated from 1977 to 1993 at the Department of General Surgery, University of Vienna, and from 1990 to 1995 at the Department of General Surgery, Hospital Lainz, Vienna. Although NPHC was diagnosed in patients ranging from 20 to 85 years of age, the median age of diagnosis was the seventh decade in women and the eighth decade in men. In our patient collective women were affected more frequently than men (79.1% versus 20.9%), a finding that concurred with published reports [1, 3–7].

Altogether 43 patients had solitary cysts, and 48 patients had polycystic disease, with cysts distributed in both lobes of the liver. Thirty-two patients (35.2%) additionally showed cystic kidneys,

Table 1. Nonparasitic cysts of the liver with regard to size and solitary cysts or polycystic disease and associated disease of the kidneys.

Cyst type	< 5 cm	5–10 cm	> 10 cm	Cystic kidneys
Solitary	10	19	14	12
Polycystic	9	24	15	20

and five had a deficit of renal function (Table 1). Pancreatic cysts were observed in seven patients, and a cyst of the spleen, lung, and ovaries was observed in one patient each.

The medical history of the patients was retrospectively examined with regard to the nature and extent of the disease, clinical symptoms, preoperative diagnosis, choice of surgical technique and subsequent complication and recurrence rates. Only one patient was lost to follow-up. The interval between the operation and follow-up examination ranged from 6 months to 17 years (mean 6.2 years). The patients were examined clinically in respect to symptoms, and a morphologic examination was performed with ultrasonography or CT. Hepatic function parameters were assessed by laboratory tests.

Results

Surgical treatment of NPHC is indicated by clinical symptoms and the size and location of the cysts as well as by the extent of polycystic disease (Table 2). The most common presenting complaint was epigastric pain (78%), followed by nausea and vomiting. The size of a solitary cyst was responsible for hepatic venous outflow obstruction in one patient. Jaundice or biliary colic was observed in 14 patients (15.4%), and cholelithiasis was the cause of disease in 8 patients. For this reason a cholecystectomy was additionally required in these eight patients and was performed laparoscopically in three. In one case of choledocholithiasis or in cases where the morphology of the NPHC was responsible for the jaundice, we always opted for the conventional surgical technique. In the presence of large cysts we decided to operate even if the patients did not have any complaints. It was done because of the high rate of complications in cases of large cysts (e.g., internal bleeding, infection, spontaneous rupture). Consequently, no surgical intervention is indicated only in patients with noncentrally located cysts measuring up to 10 cm and no or only minimal symptoms (n = 7). This subgroup of patients did not require surgery even at a later point in time.

The use of *CT-guided aspiration* is technically limited to small cysts. It was performed in nine patients and led to recurrent symptoms in seven within a short period. These patients then underwent conventional surgical fenestration, which was successful in all cases.

Laparoscopic fenestration with widest possible excision of the cystic wall was performed in five patients with cysts measuring up to 10 cm in diameter. None of these patients had recurrent symptoms. Only one patient had a solitary cyst. In three cases of coexistent cholecystolithiasis, the cyst operation and cholecystectomy were performed synchronously. Laparoscopic fenestration was successfully performed three times in five patients with cysts measuring more than 10 cm. One woman with a solitary cyst underwent a second laparoscopic fenestration because of recurrence. One woman was initially operated by the laparoscopic technique, but the operation was interrupted because the patient

Table 2. Therapeutic procedures in patients with nonparasitic cysts of the liver.

Procedure	Solitary cysts	Polycystic disease
CT puncture	7 (5)	2 (2)
Laparoscopic fenestration	5 (1)	5
Conventional fenestration	20	34 (7)
Liver resection	4	5
Cystojejunostomy	3	0

Numbers in parentheses represent patients who developed recurrent symptoms.

developed bleeding that was not staunchable by endoscopic means. Another woman with multiple, large cysts was not completely free of symptoms after surgery but was not exposed to any further surgical treatment because imaging studies showed that the cysts had been emptied. Hence further surgery was deemed unnecessary.

Eleven patients with cysts measuring < 5 cm were treated by the *conventional surgical technique*. In cases of solitary cysts (n = 4) the indication for conventional surgery was the presence of symptoms caused by a central location (jaundice). In the remaining seven patients the indication for surgery was the presence of multiple, symptomatic cysts. None of these patients had a recurrence. Altogether 43 patients with cysts measuring 5 cm or more were treated by conventional surgical fenestration or excision. Of 27 patients with multiple cysts, 7 (26%) required a second intervention. In contrast, patients with solitary cysts never developed recurrent symptoms.

A cystojejunostomy was performed in three patients in whom the cysts were found to have bile content and cystobiliary connection. All cysts in this group were solitary, with two of them larger than 10 cm. One patient with a medium-sized cyst underwent homologous liver transplantation 6 months after cystojejunostomy due to a centrally located cholangiocellular carcinoma. The further course of disease was uneventful.

Partial hepatectomy was performed in nine patients with no complications. Five patients had multiple cysts. All patients had cysts with a largest diameter of 5 cm or more. Four patients with solitary cysts in the left lobe of the liver underwent left lobectomy. One patient underwent left hemihepatectomy, two right hepatectomy, and one right lateral segmentectomy. A mean of 2.6 hepatic segments were resected in cases of polycystic disease. Cysts that were found in other sections of the liver were subsequently fenestrated if they were larger than 2 cm. Long-term results were excellent in these patients. All patients were free of recurrences.

Infectious complications were observed in four patients. Two patients developed hepatic abscesses that finally led to death (one of these patients had undergone laparoscopic surgery), and two patients developed a subphrenic abscess that was aspirated under CT guidance. One patient who had undergone conventional surgical fenestration developed postoperative bleeding, because of which a right lateral segmentectomy was performed. One patient who developed bile leakage had to be reoperated.

Especially in cases of cysts measuring > 5 cm, we frequently observed postoperative ascites that necessitated a prolonged period of drainage. Only one patient with a cyst > 10 cm had constant subjective complaints due to persistent ascites after removal of the drain.

Twenty patients (22%) died during the period of observation, of whom 15 were women and 5 were men. As mentioned earlier, two patients expired during the immediate postoperative period because of septic complications.

A remarkable aspect of the patients who expired was the frequent presence of malignancy (of the lung, ovary, breast, stomach, and colon). A total of 11 patients (12%) died because of malignancies. Three other patients with Potter's III disease died of septic complications during the course of the basic disease.

Discussion

We have summarized our experience with the spectrum of therapeutic procedures for treatment of NPHC. Our aim was to elucidate the advantages and disadvantages of the various methods. As mentioned earlier, *CT-guided aspiration* of small cysts is technically difficult, and the indication for this type of therapy is not unequivocal. Cysts larger than 5 cm in diameter can be easily treated with CT-guided aspiration, but the rate of recurrence is so high that this procedure is not advisable except for diagnostic purposes. Guided aspiration is frequently combined with the use of a sclerosing agent such as fibrin adhesive, electrocoagulation, alcohol, or tetracycline [2, 5, 8–12], but this mode of treatment was not used in any of our patients.

Our findings corroborate those of studies performed on larger series of patients, which indicate that percutaneous aspiration can be regarded only as a palliative measure [1, 3–6, 9, 13–15]. Moreover, it could lead to severe complications if the gallbladder or colon is inadvertently perforated [2]. Hence the only indications for this procedure are either very high operative risk or the need to perform a cytologic diagnosis.

Laparoscopic surgery (which was used in 10 of our patients) is a new, much less stressful but equally effective technique. The few published reports of this technique are anecdotal in nature [9, 16-19]. Recently reports of larger series have demonstrated its effectiveness for solitary cysts and even polycystic liver disease [20, 21]. However, recurrent symptoms were observed in 23% and 61% of patients, respectively. Alternatively, a simple cyst can be removed by a GIA clip suture device [22]. Although laparoscopic fenestration has been primarily used for treatment of solitary cysts [7, 20], we used this technique to treat patients with multiple cysts of every size, and none of these patients suffered a relapse (n = 5). However, of the five patients with solitary cysts, one developed recurrent symptoms necessitating repeat laparoscopy. The second intervention was successful in this patient. In another case laparoscopic surgery could not be concluded because the patient developed a hemorrhage of the cyst wall. Obviously, a decisive criterion of success of the laparoscopic technique is the widest possible excision of the cystic wall to avoid postoperative adhesions, which cause the cysts to be refilled. This principle generally applies to both laparoscopic and conventional surgical techniques. A recurrence rate of 11% was registered in nine patients who were finally operated by the laparoscopic technique. Although general clinical experience with this technique is limited, this recurrence rate is sufficiently low to justify recommendation of this technique as a routine procedure. In an earlier study comprising an even smaller group of patients [23] we found a higher recurrence rate for laparoscopy than for the conventional surgical technique. This proportion has now been reversed (11% vs. 13%).

Excision of the cyst has been propagated as the method of

choice for *conventional surgical therapy* because it is associated with low recurrence rates [1, 3–5, 11, 13, 24], although fenestration with widest possible excision of the cystic wall may now be regarded as an equally effective technique and is given particular preference in cases of polycystic disease [14, 15, 25–28]. The recurrence rate for this technique increases with increasing rigidity of the cyst wall [25].

Excision has been used as the primary technique in cases of small peripheral cysts [1, 3–5, 13, 29]. Although the recurrence rate for conventional surgery (13%) is well within acceptable limits, we identified a subgroup of patients who were at high risk for recurrence. All recurrences were observed in patients with multiple cysts measuring 5 cm or more. Of the 27 patients of this group, 26% developed recurrences that required surgical treatment. Owing to this relatively high rate of recurrence, conventional surgery cannot be recommended for these patients. On the other hand, it is remarkable that none of the four comparable patients who were treated laparoscopically had recurrences. According to our data laparoscopic fenestration is a useful therapeutic alternative in these cases, whereas others recommend caution using this technique in patients with polycystic disease [21]. Apart from less surgical stress to the patient, the laparoscopic technique did not prove to be significantly more advantageous than conventional surgery, a fact that could be attributed to the small number of patients treated by laparoscopy.

Partial hepatectomy with fenestration of the cyst, a technique we used in five patients with multiple cysts, was successfully performed without recurrence of symptoms. A combination of partial hepatectomy and fenestration of the cyst has been frequently used in cases of polycystic enlarged livers [26, 30-32]. In keeping with these results, we found this procedure to be indicated in a select group of patients with polycystic disease, especially in cases where the liver is massively enlarged and fenestration of the cyst does not help reduce the size of the liver. In cases of NPHC, hepatectomy can be performed with a low rate of morbidity and practically no recurrence [26, 30–33]. Only in one study has a recurrence rate of 25% been reported [26]. Partial hepatectomy for surgical cleaning of the NPHC is particularly suitable in cases of large, solitary cysts in the left lobe of the liver. A technically simple lobectomy can help remove the entire cyst with practically no recurrence. Morbidity has also been low with this technique [1, 3, 13, 26, 29].

Four patients from our series who had undergone left lobectomy of the left lobe were subsequently symptom-free. This result should be viewed in the context of the fact that large solitary cysts are generally associated with a low rate of recurrence. Hence in the presence of this morphology we primarily recommend laparoscopic surgery, provided the patient does not present with additional complications that necessitate laparotomy. Solitary cysts containing bile were treated with cystojejunostomy in three cases. One patient with multiple cysts with a cystobiliary connection underwent partial hepatectomy. No complications were encountered in these patients. Cystojejunostomy is an excellent alternative for this indication [4, 5, 14, 15, 29], although the possibility of secondary infection from the cystojejunostomy should be considered [14]. In cases of multiple cysts, the indication for cystojejunostomy or partial hepatectomy should be established individually.

Because of the frequent occurrence of postoperative ascites, we refrained from performing prolonged external drainage. Although

drainage of this type has been reported in the literature [1, 4-7], it is generally not recommended. On the other hand, some authors recommend suppression of ascites with somatostatin or H_2 -blockers [6, 7]. We did not use these forms of therapy, but we did perform routine postoperative drainage. With one exception, all drains were removed easily.

Infection has been frequently observed in large series of patients and has led to death in some cases [3–5, 29]. Possibly these deaths were also related to an infected cystic content. There are no reports on the concomitant occurrence of malignancy in patients with NPHC, probably because small numbers of patients were studied and the periods of observation were relatively short.

A standardized therapeutic procedure cannot be recommended for NPHC. In cases of asymptomatic cysts measuring > 10 cm, the complication risk due to secondary infection, spontaneous perforation, or internal bleeding is rather high. These patients should be treated by surgery [4, 5, 29]. Surgery is also indicated in cases of solitary, small, centrally located cysts because of the risk of local compression on the hilar structures.

As primary intervention, we recommend laparoscopic surgery with the widest possible fenestration of the cyst wall in all cases of NPHC. The advantage of this procedure is (1) low stress for the patient and (2) a recurrence rate similar to that for open fenestration. Partial hepatectomy is an equally good surgical technique, especially in patients with large, polycystic livers in whom simple fenestration would not provide any relief and would not adequately reduce the size of the liver.

For large solitary cysts that occupy segments II and III, partial hepatectomy has been successful, and the morbidity has been acceptably low. Nevertheless, for primary treatment of this indication, we recommend laparoscopic surgery because it is less stressful for the patient.

Résumé

La nature et le mode de survenue, et par conséquence, l'attitude thérapeutique des kystes non parasitaires du foie (KNPF) sont très variables. A partir de notre expérience, nous avons essayé de trouver l'attitude la mieux adaptée à chaque patient. Nous avons analysé rétrospectivement les dossiers de 91 patients ayant un KNPF, traités de manière chirurgicale entre 1977 et 1995. Les KNPF non symptomatiques, périphériques, inférieurs à 10 cm de diamètre, ne nécessitent pas de traitement. La ponction guidée par tomodensitométrie (TDM) (n = 9) doit être considérée comme une méthode tout à fait palliative en cas de kystes symptomatiques. En effet, sept patients traités ainsi sont redevenus symptomatiques très peu de temps après leur ponction. Le traitement standard de ces kystes est la fénéstration comprenant une excision aussi large que possible de la paroi kystique. Cette intervention peut être accomplie soit par laparoscopie (n = 10), soit par chirurgie conventionnelle (n = 54). Un patient, opéré initialement par laparoscopie, a du être converti en raison d'une hémorragie. Trois patients ont eu une cholécystectomie sous laparoscopie en mēme temps. Le taux de récidive était similaire: 11% dans le groupe laparoscopie vs 13% dans le groupe conventionnel. Le traitement conventionnel a toujours été un succès en cas de chirurgie des kystes solitaires. Cependant, dès lors qu'il existe plusieurs kystes, mesurant plus de 5 cm de diamètre, la chirurgie conventionnelle a été suivie, chez 26% des patients (7/27), de récidive de symptomes qui ont ensuite nécessité une

deuxième intervention. La résection partielle du kyste (n = 9) a été un succès dans le cadre de la maladie polykystique (n = 5) associée à une hépatomégalie, ainsi qu'en cas de kystes volumineux solitaires du lobe gauche (n = 4). Chez les patients ayant une communication biliaire (n = 3), nous avons effectué une kystojéjunostomie. Globalement, le taux de complication était bas. En dehors de l'ascite postopératoire, ne nécessitant aucun traitement ultérieur, des complications infectieuses ont été observées chez quatre patients. Vingt patients (22%) sont décédés pendant une période moyenne de 6.2 ans. Le décès était fréquemment associé à une maladie maligne (11/20). Après fénéstration de kystes multiples dont le diamètre dépasse 5 cm, il existe un risque éiveé de récidive. Parml ces patients, lorsqu'il existe une maladie polykystique et une hypertrophie du lobe gauche, la fénéstration simple ne suffit pas et la résection partielle du foie est une excellente alternative thérapeutique. Les résultats de cette étude montrent que la fénéstration sous laparoscopie devrait remplacer la technique conventionnelle comme technique de base en cas de KNPF en raison de sa moindre agressivité et son taux de succès similaire.

Resumen

Los quistes no parasitarios del hígado (QNPH) son de gran variabilidad en cuanto a apariencia y aproche quirúrgico. El tratamiento varía según la naturaleza y la apariencia de la enfermedad. En el presente estudio se discuten las modalidades óptimas de tratamiento para las diversas formas de QNPH, con fundamento en la naturaleza variable de la enfermedad y las diferentes opciones terapéuticas. Se hizo el análisis retrospectivo de 91 pacientes con QNPH sometidos a tratamiento quirúrgico entre 1977 y 1995. Los quistes periféricos asintomáticos que miden hasta 10 cm no requieren tratamiento. La aspiración guiada por TAC (n = 9) debe ser considerada como una medida paliativa; en un corto intervalo, el método dio como resultado recurrencia de la sintomatología en 7 de nuestros pacientes. El tratamiento estándar de los QNPH es la fenestración con la más amplia resección posible de la pared del quiste, lo cual puede ser realizado por vía laparoscópica (n = 10) o por cirugía convencional (n = 54). Un paciente fue intervenido inicialmente por la técnica laparoscópica pero presentó sangrado, lo cual requirió conversión al método abierto. Tres pacientes recibieron colecistectomía laparoscópica concomitante. Las tasas de recurrencia fueron similares: 11% en el grupo de la laparoscopia vs. 13% en el de cirugía convencional. El tratamiento quirúrgico convencional fue siempre exitoso en los casos de quistes solitarios. Sin embargo, en los casos de quistes múltiples, resultó en recurrencia de los síntomas en 26% de los pacientes (7/27), quienes requirieron una segunda operación. La resección parcial del hígado (n = 9) fue practicada con éxito en casos de enfermedad poliquística (n = 5) con aumento concomitante en el tamaño del hígado, así como en casos de grandes quistes solitarios del lóbulo izquierdo del hígado (n = 4). En los pacientes en quienes encontramos que los quistes se comunicaban con el sistema biliar (n = 3), practicamos una cistoyeyunostomía para drenaje de la bilis. La tasa de complicaciones fue baja. Además de frecuente ascitis postoperatoria que no requirió intervención, observamos complicaciones infecciosas en 4 pacientes. Veinte (22%) murieron durante un período medio de seguimiento de 6.2 años. Como dato interesante, las muertes aparecieron comúnmente asociadas con neoplasias malignas (11/20). Luego de la fenestración de quistes múltiples mayores de 5 cm, los pacientes quedaron con alto riesgo de recurrencia. Esta es la razón por la cual la resección parcial del hígado es una excelente alternativa terapéutica en pacientes seleccionados con enfermedad poliquística y hepatomegalia masiva, en quienes la enfermedad no puede ser controlada mediante simple fenestración. Los resultados del presente estudio demuestran que la fenestración laparoscópica debe reemplazar a la técnica quirúrgica convencional como el patrón oro en casos de QNPH, por cuanto es menos traumática para el paciente y se asocia con una tasa de éxito similar.

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Invited Commentary

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The recognition of NPHC has increased with the improved accuracy of hepatic imaging. Koperna et al. call attention to the clear need for clinicians to address the management of patients with NPHC rationally and efficaciously. They relate their experience with NPHC developed over nearly two decades. Importantly,

their mean follow-up after intervention was more than 6 years. What are the key lessons learned? (1) Aspiration alone was condemned to failure. (2) Laparoscopic fenestration was effective and safe. (3) Open fenestration with or without resection is the best approach for multiple cysts, cysts with complications, or polycystic liver disease. Their final therapeutic algorithm favors initial laparoscopic management succeeded by open laparotomy in cases of failure, complications, or polycystic disease. Despite this large experience, answers to several questions are required before their guidelines can be widely adopted.

First, should asymptomatic cysts be selectively observed (<10 cm) or selectively treated (>10 cm)? Data herein and elsewhere are sparse. Whether intrahepatic location (peripheral or deep),

size, number, or rate of growth leads to symptoms or complications is unknown. No prospective data substantiate significant risk for observation of cysts regardless of size. Safe treatment does not necessarily justify intervention. Second, image-guided aspiration alone appropriately is condemned, but sclerosis was not employed herein. Although reports in the radiologic literature are encouraging, follow-up is limited. If aspiration with sclerosis proves durable, the role of the laparoscopy would necessarily be deferred. Third, Koperna et al. advocate a primary role for laparoscopy in patients with NPHC except for polycystic liver disease. However, others have identified limits for the laparoscopic approach. Cysts in segment 7 and 8 or cysts whose surface area adjacent to the liver capsule is small may be better served by laparotomy. Careful selection criteria can reduce recurrence and should be recognized. Finally, laparoscopy has no current role in the treatment of complex polycystic liver disease. The massive hepatomegaly associated with architectural distortion of vascular and biliary structures, large intrahepatic vascular collaterals, and often dense adhesions to surrounding organs present prohibitive hurdles for effective hepatic volume reduction.

The trend toward a primary laparoscopic approach is reasonable. However, further experience and follow-up and comparisons to alternatives are necessary before laparoscopic approaches become the treatment of choice for NPHC.