

Case Report

Percutaneous transhepatic electrohydraulic lithotripsy for intrahepatic bile duct stones after choledochal cyst excision

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Excision and hepaticojejunostomy are well-established treatments for choledochal cysts. However, the formation of intrahepatic bile duct stones has been reported as one of the most serious long-term complications on follow-up of choledochal cyst excision. The reported incidence of this complication varies from a small percentage of patients to 10% or more. Various procedures have been reported as treatments for postoperative bile duct stone formation. We report a case in which percutaneous transhepatic electrohydraulic lithotripsy (EHL) was used in the treatment of bile duct stones that developed after choledochal cyst excision. A 17-year-old boy, who had undergone choledochal cyst excision and hepaticojejunostomy when he was 27 days old, presented with colic abdominal pain in the right hypochondriac region and liver dysfunction. CT performed in our emergency department revealed intrahepatic bile duct stones in minimally dilated intrahepatic bile ducts in both lobes. Percutaneous transhepatic cholangiography showed packed stones distal to the right hepatic duct and stenosis of the right hepatic duct; there was no evidence of stenosis at the hepaticojejunostomy anastomosis. He underwent extracorporeal shockwave lithotripsy under general anesthesia. Although the large stone was fragmented into smaller stones, they were not small enough to be eliminated due to stenosis of the hepatic duct. After balloon dilatation of the stenosis, the patient underwent percutaneous transhepatic EHL. The fragmented stones were then thoroughly washed out from the intrahepatic bile duct. EHL is an effective and less invasive treatment for intrahepatic bile duct stones after choledochal cyst excision.

ACTA MEDICA NAGASAKIENSIA 56: 99 - 102, 2011

Keywords: Choledochal cyst, percutaneous transhepatic electrohydraulic lithotripsy, intrahepatic bile duct stones, extracorporeal shockwave lithotripsy

Introduction

Excision and hepaticojejunostomy are well-established treatments for choledochal cysts. However, formation of intrahepatic bile duct stones has been reported as one of the critical, long-term complications on follow-up.¹⁻⁵ The reported incidence of postoperative intrahepatic bile duct stone formation varies from a small percentage of patients to 10% or more,¹⁻⁵ and various procedures have been reported

as treatments for this condition.^{2,6-10} We report herein a case in which percutaneous transhepatic electrohydraulic lithotripsy (EHL) was effective in the removal of bile duct stones.

Case report

A 17-year-old boy, who had undergone choledochal cyst excision and hepaticojejunostomy when he was 27 days old,

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Received March 10, 2011; Accepted April 18, 2011

presented with colic abdominal pain in the right hypochondriac region and liver dysfunction. He was transferred to our hospital for further examination. Aspartate transaminase, alanine transaminase, γ GTP and C-reactive protein were elevated to 86 IU/l, 157 IU/l, 425 IU/l and 2.71 mg/dl, respectively. Computed tomography (CT) showed intrahepatic bile duct stones in minimally dilated intrahepatic bile ducts of both lobes (Figure 1). Percutaneous transhepatic cholangiography of the right hepatic duct revealed right hepatic duct stenosis distal to the packed stones (Figure 2). No stenosis was apparent at the hepaticojejunostomy anastomosis. Initially, we performed extracorporeal shockwave lithotripsy (ESWL) for the removal of the stones. The procedure was somewhat effective in fragmenting large stones into smaller pieces; however, the pieces were not small enough to be eliminated due to hepatic duct stenosis. We then applied balloon dilatation to the stenosis and used percutaneous transhepatic cholangioscopy to remove the stones by mechanical lithotripsy (ML) through



Figure 1. Preoperative computed tomography showed multiple stones incarcerated in the right lobe with bile duct dilation (A & B, white arrowheads) and in the left lobe (C, black arrowheads)



Figure 2. Percutaneous transhepatic cholangiography showed multiple stones in the right lobe proximal to stenosis of the right bile duct (white arrowhead). A black arrowhead indicates the hepaticojejunostomy.

a flexible choledochoscope (CHF Type T20; Olympus, Tokyo, Japan) under general anesthesia. This method was also somewhat effective; however, due to the amount of packed stones, there was insufficient working space. We then performed cholangioscopy-directed EHL. The generator used for the procedure was a Storz Calcutript 27080 (Karl Storz GmbH & Co. Kg, Tuttlingen, Germany) (Figure 3). The stones were easily fragmented by bringing the EHL close to the surface of each stone and carefully generating shock waves. The stones were then washed away with saline (Figure 4). We applied this method to each hepatic lobe at 1-week intervals. A total of 500 shocks were used to fragment the stones for two days. Postoperative radiological investigation showed a small, residual stone in the right lobe (Figure 5). Although the patient suffered from iterative cholangitis 12 months after the procedure, there was no regrowth of the intrahepatic duct stone. He is currently doing well and has not had any recurring episodes of bile duct stones.



Figure 3. An electrohydraulic shock wave generator (Storz Calcutript 27080) for EHL.

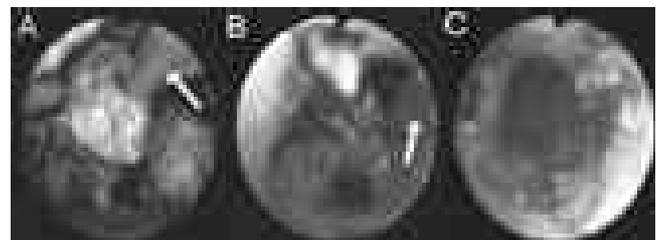


Figure 4. Cholangioscopic view showed intrahepatic bile duct stones before EHL. A white arrow indicates the tip of the EHL probe (A). After several shots of EHL, the stones were fragmented (B). The fragments were washed away with saline after EHL. Post-procedure cholangioscopy showed that fragments had been cleared from the intrahepatic bile duct. There were minor mucosal damages of the intrahepatic bile duct (C).



Figure 5. Postoperative computed tomography showed a small residual stone in the left intrahepatic duct (white arrow) and a percutaneous transhepatic cholangiodrainage tube in the right intrahepatic duct.

Discussion

The development of intra- and extrahepatic bile duct stones has been reported as one of the most serious complications after choledochal cyst excision.¹⁻⁵ Intrahepatic bile duct stone formation is thought to be caused by bile congestion due to an anastomosis stricture from a hepaticoenterostomy or congenital intrahepatic duct stenosis.^{1,5}

Various treatments have been reported for intrahepatic bile duct stone formation after choledochal cyst excision.^{2,6-10} Although liver resection and modification of hepaticojejunostomy result in a higher clearance of stones, such invasive surgery should be avoided in younger patients. A less invasive approach is ESWL, which is performed without a laparotomy or puncture. Okada et al. reported excellent results using this method for intrahepatic bile duct stone after choledochal cyst resection², but in the present case we did not obtain good result. Using ESWL, we were unable to maintain focus on the intrahepatic duct stones, as the shock waves caused them to move. This approach, which is a less invasive method, is useful only in special cases-e.g., impacted intrahepatic bile duct stones or immobile stones such as ureteral calculi. Intracorporeal techniques such as ML, EHL, and laser lithotripsy are also options for fragmentation of impacted stones in the intrahepatic ducts. These intracorporeal procedures require continuous visualization of the fragmentation process with a peroral mother-baby cholangioscopy system or percutaneous transhepatic cholangioscopy. Patients with stone formation require a percutaneous transhepatic approach (PTA) or a trans-Roux-en-Y (RY) limb approach

with a laparotomy due to Billroth II reconstruction and bilioenteric anastomosis. Relaparotomy is also an option; however this could lead to bowel injury during adhesiolysis as well as difficulty in identifying the RY limb. In contrast, PTA offers the possibility of a repeated procedure without the need for a laparotomy.¹⁰ We chose PTA and EHL in the present case, as this patient had numerous stones impacted in the bilateral intrahepatic bile duct and stenosis of the right hepatic bile duct. The ML approach, introduced by Demling et al. in 1982, has also shown effective results in the removal of difficult bile duct stones.¹¹ However, it has been reported that the ML basket could not capture stones exceeding 3 cm.¹² In addition, the basket was unable to open due to multiple impacted stones that clogged the entire bile duct.¹³ Originally used in urological procedures, EHL produces a high-voltage discharge that creates high-frequency, hydraulic pressure waves. In 1987, Ligoury et al. reported the first case of EHL treatment in a patient with common bile duct stones.¹⁴ The EHL probe must be placed on the surface of the stones and away from the bile duct wall, as the pressure waves can damage normal tissue. Bile duct perforation and bleeding have been reported after EHL because of direct contact between the EHL probe and bile duct wall.¹⁴⁻¹⁶ Thus, during EHL in the present patient, the stones were directly visible under a continuous flow of normal saline. Fragmented stones were then washed out through the RY limb or the PTA route. EHL has been found to be an effective treatment for the fragmentation of intrahepatic bile duct stones, with a success rate of 80-100%.¹⁷⁻¹⁹

Stenosis and repeated cholangitis of the intrahepatic bile duct can lead to bile stasis and intrahepatic stone formation, which may be a high-risk factor for carcinogenesis.^{20,21} We believe that cholangioscopy-directed EHL is an effective and alternative method for removing intractable intrahepatic bile duct stones which developed after choledochal cyst excision.

Conclusion

ESWL and ML or EHL with a PTA approach are effective and less invasive procedures for treating intrahepatic bile duct stone formation after choledochal cyst excision. An EHL with a PTA approach is an effective procedure for the fragmentation of intrahepatic bile duct stones when ESWL or ML do not work and offers the possibility of a repeated procedure without the need for a laparotomy.

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