CASE REPORT

Acute biliary pancreatitis and cholecystolithiasis in a child: One time treatment with laparoendoscopic “Rendez-vous” procedure

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INTRODUCTION

Acute biliary pancreatitis (ABP) is rare in childhood and endoscopic sphincterotomy should be avoided in the child due to the risk of both early and late complications but, when necessary, the optimal timing between endoscopic procedure and cholecystectomy is still uncertain. In the child the literature concerning therapeutic ERCP with ES for ABP is scarce, and ERCP, combined with ES or not, carries also the well known risk of iatrogenic acute pancreatitis. The optimal diagnostic and therapeutic approach and the timing of the procedures in children with cholecystolithiasis will often depend on the level of expertise of both the biliary endoscopist and laparoscopist.

A nine years old child with acute biliary pancreatitis underwent the first successful laparo-endoscopic “Rendez-Vous” procedure in which endoscopic drainage of the common bile duct and laparoscopic cholecystectomy were performed simultaneously. The favourable outcome of the reported case led us to deepen this issue.

CASE REPORT

A nine-year-old girl, weighing 52 kilograms, BMI of 27, was urgently admitted to our department because of pain in the right upper abdomen associated with vomiting. The only abnormal laboratory tests were the serum ALT 163 u/L (normal value 10-55), amylase 131 U/L (normal value < 110), lipase 482 U/L (normal value < 300), and ALP 168 U/L (normal value 38-126). Abdominal ultrasonography revealed multiple gallstones (3-5 millimeter) and sludge in the gallbladder and a normal common bile duct (CBD). The patient was kept NPO with intravenous fluids and analgesics and was discharged from the hospital after being asymptomatic for 24 h against medical advice, because her parents refused laparoscopic cholecystectomy (LC). The patient was followed in the paediatric outpatient clinic where metabolic or haematologic causes of cholecystolithiasis were excluded.

Three weeks later the patient was again urgently admitted because of a recurrence of abdominal pain, vomiting and a fever (38.6°C). Ultrasonography showed gallstones, a dilated CBD (8 mm) and pancreatic edema. Laboratory test showed a nine fold increased serum amylase of 998 U/L and of serum lipase 2430 U/L (normal value < 300), and ALP 168 U/L (normal value 38-126). Abdominal ultrasonography revealed multiple gallstones (3-5 millimeter) and sludge in the gallbladder and a normal common bile duct (CBD).

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value 1.3 and 0.5) respectively and other pathologic parameters were: gamma GT 479 U/L (normal value 78), ALT 1032 u/L, AST 584 U/L (normal value 10-45), LDH 1855 U/L (normal value 313-618), ALP 283 U/L and C reactive protein (CRP) 6.8 mg/dl (normal value <1). White blood cell count (WBC) was 13,200 (normal value 6000-9000) with 91% polymorphonucleates, and blood glucose was 132 mg/dL. According to the Atlanta classification and Ranson’s criteria\(^1\) the diagnosis of mild acute biliary pancreatitis was given and a conservative treatment was started with fluids, analgesics and imipenem. The main symptoms disappeared after 12 h so that the conservative treatment was continued. After 48 h a CT scan showed a pancreatic and peripancreatic oedema that was classified as Balthasar stage B\(^1\). The bilirubin remained slightly abnormal (1.62 total and 0.74 direct) but serum amylase and lipase were increased to 1300 and 30560 U/L respectively, whereas CRP reduced to 4.4 mg/dL. A cholangio-MRI was performed (Figure 1) showing a filling defect in the upper portion of the CBD that was highly suspicious for a gallstone, sludge or mucinous aggregate.

Based on increased experience in adults with cholecystocholedocolithiasis, we decided to perform a laparoendoscopic “Rendez-vous” for both the endoscopic drainage of the CBD and treatment of gallstones despite the fact that there was no evidence in the literature of this procedure being performed on a child that was considered suitable, without contraindication and useful. Laparoendoscopic “Rendez-Vous” procedure was as follows: The whole laparo-endoscopic procedure was carried out to maintain the patient in the same supine “french” position used for the laparoscopic cholecystectomy. Three trocars (10-10-5 mm) were introduced with open technique. The first manoeuvre was the preparation, encircling and clipping of the cystic duct to avoid migration of stones in the CBD. An intra-operative cholangiography was performed to introduce a Pedinelli catheter (8 fr) in the cystic duct. This showed a dilated CBD and a delayed passage of the contrast medium in the duodenum (Figure 2), but could not confirm the filling defect. The X-ray picture of the whole biliary tree remained unmodified without any flow of the contrast medium in the duodenum, up to 30 minutes after the injection and despite intravenous administration of a dose of antispastic drug (cymetropio bromide). This led us to suspect an underlying sphincter of Oddi dysfunction but a manometry was considered unnecessary because the indication for retrograde exploration of the papilla was given. During the time waiting to control the passage of the contrast medium in the duodenum, the cholecystectomy was carried out. The endoscopist performed the procedure using an adult lateral view duodenoscope. The papilla appeared normal but without any flow of bile. A guide wire could be introduced through the papilla and its correct position inside the still contrasted CBD could be confirmed. In this way the standard and more dangerous unpreventable retrograde injection of the Wirsung duct could be avoided. A paediatric endoscopic balloon catheter Olympus Swift ® (4 mm diameter) was then introduced in the CBD, inflated, and retrieved trough the papilla. Some biliary sludge could be retrieved and immediately bile flowed. This procedure was repeated twice and a gentle dilatation of the papilla was also performed for 2 minutes. The endoscope was then extracted and the laparoscopic procedure ended after clipping of the cystic duct. The post-operative laboratory controls showed the normalization of amylase, lipase and bilirubin the day after. On day two after the procedure a control US showed a normal CBD. The patient was discharged three days after the procedure. Neither recurrence of symptoms, nor abnormality in laboratory tests or US controls occurred up to the last 24 mo follow-up.

**DISCUSSION**

Acute biliary pancreatitis in the child is rare and the main causes are congenital disorders or malformations of the pancreatic-biliary ductal system. In children, the main indication of diagnostic ERCP with or without endoscopic sphincterotomy (ES) are biliary atresia, intrahepatic cholestasis, anomalous arrangement of the pancreatic-biliary ductal system including the dilatation of the CBD\(^3,4\). In the adult, ES is a part of the treatment of ABP when this is caused by stones or sludge impacted in the papilla.\(^6,7\) Concerning the timing of ERCP +/- ES, early ap-
approach is not more beneficial than conservative treatment in patients without jaundice. In the child the literature concerning therapeutic ERCP with ES for ABP is scarce, especially because the pancreatitis, according to Hsu and Novak, is often not “biliary” in the “adult” meaning of the term. In the reported case the pancreatitis was truly “biliary”, because related to micro-lithiasis and sludge and also combined with an underlying sphincter of Oddi dysfunction which can often be related to recurrent pancreatitis also in children. The treatment of ABP in childhood needs a multidisciplinary approach to optimize laparoscopic treatment, and intraoperative cholangiography is considered mandatory to minimize unnecessary ERCP. In fact ERCP, combined with ES or not, carries the well known risk of iatrogenic acute pancreatitis because of papilla manipulation, pancreatic intraductal hyperpression and/or chemical damage all factors related to unpreventable retrograde injection of the pancreatic duct. The sole case of urgent ERCP-ES in a 9-year-old patient with ABP due to a stone impacted in the papilla was reported recently and was performed under anaesthesiological control of sedation. This patient was discharged after ES, with a planned laparoscopic cholecystectomy and was therefore still at risk of recurrent pancreatitis in the possible event of an incomplete sphincterotomy. There is the well known problem of the optimal timing between ERCP and LC, also considered “The bilateral interface…” between the two procedures but the optimal diagnostic and therapeutic approach in children will often depend on the level of expertise of both the biliary endoscopist and laparoscopist. All these considerations and the favourable outcome of the case convinced us that simultaneous laparoendoscopic treatment carries great advantages especially in children. The literature shows that comparable good success rate of ERCP between adults and children (98% vs 97.5%) with a comparable low incidence of complications (2.5% vs 3.4%) are obtainable only by very experienced teams but unfortunately complications can reach up to 33%. Therefore ERCP in the paediatric population should be minimized because stones often pass spontaneously and also because the only sure way to avoid post-ERCP complications is to avoid ERCP itself. This is certainly possible with the combined laparo-endoscopic approach because ERCP is avoided and ES is limited only to those patients with stones or abnormal cholangiography that cannot be resolved laparoscopically via the cystic duct or when a sphincter of Oddi dysfunction can not be resolved through a simple dilatation. The balloon papilla dilatation carries comparable effectiveness but with a discordant incidence of relevant complications like pancreatitis that can be related to unnecessary retrograde injection of the pancreatic duct or manipulation, but anyway in children dilatation should be preferred to preserve the papilla. The dilatation is also effective in early stone retrieval during the attack of ABP.

Other favourable considerations concerning effectiveness of this combined approach can be found in the literature concerning the standard sequential approach. In a large prospective randomized study the effectiveness of ERCP/ES to clear CBD stones was only 84%, when performed prior to laparoscopic cholecystectomy and therefore 16% can be ineffective. Unfortunately, these patients often also have to undergo urgent surgery, with exposure to a second X-Ray for the mandatory cholangiography and often the need for a surgical opening of the CBD. This carries the need of relevant surgical skill but carries also an increased risk of complications, often need of T tube or other biliary drain, and prolonged hospitalization. In the child both open and laparoscopic surgery and especially suturing of the small duct requires a high degree of surgical expertise to avoid the risk of late stenosis of the CBD. On the other hand, laparoscopic clearance of CBD stones in a large prospective study of adults was impossible in 15% of the cases, despite all being operated on by experienced surgeons, and conversion to open surgery or post-operative ERCP carried a relevant incidence of biliary complications, but the post-operative ERCP can itself be related to risk of ineffective.

A recent analysis of management of suspected common bile duct stones in children shows clearly the poor results and pitfalls of the two common pathways of the sequential treatment with ERCP-ES and laparoscopic cholecystectomy. If performed prior to cholecystectomy a total of 71% of ERCP were unnecessary and 7% failed, if performed after cholecystectomy 50% of ERCP were unnecessary because stones were not found. Despite a reported incidence of 0% complication rate, 9 of 12 ES (75%) were unnecessary based on findings of an intra-operative cholangiography, some days prior. Therefore, a large number of unnecessary invasive procedures, all potentially related to morbidity and mortality, are often ineffective especially because they are out of synch with the evolving pathophysiology of gallstone disease. On the other hand, the simultaneous approach of RV is tailored to a “real time” picture of the CBD and the compliance of the papilla, eliminating all previous and subsequent unnecessary procedures, and also minimizing the factors of ineffectiveness. Concerning ABP, endoscopy and laparoscopy, a recent review pointed out that the sequential approach of ERCP-ES first followed by LC is the least efficient, and despite simultaneous treatment is theoretically the best option, the laparoscopic CBD clearance is considered difficult and time consuming. The main experiences with the simultaneous laparoendoscopic approach were extremely positive, showing a high effectiveness in stone clearance of 97%-100%, a low morbidity and hospital stay comparable to simple laparoscopic cholecystectomy. It is also important to consider that for a child younger than 10 years of age the ERCP usually requires general anaesthesia and an additional administration is certainly needed for the successive cholecystectomy. On the other hand, to perform the laparoendoscopic RV only one administration of anaesthesia and related psychological trauma is necessary for the child. The outcome of the case and the analysis of the literature, with a comparison of combined laparoendoscopic Rendez-Vous to the other available options convinced us that considering the technical aspects, the related risk of iatrogenic complications, optimization
of the timing and the effectiveness of the procedure, the laparo-endoscopic Rendez-Vous in these cases is the best option for the child, perhaps also for the adult, the surgeon, the endoscopist and the hospital itself.

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