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The benefit of laparoscopic cholangiography in differential diagnosis of biliary atresia and a review of the literature

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ABSTRACT

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Keywords:

Biliary atresia Kasai Laparoscopic cholangiography Pediatric cholangiography (LC) and to compare the open and LC. LC's performed in our clinic between 2008-2013 for differential diagnosis of biliary atresia (BA) in cases of prolonged jaundice were evaluated retrospectively and compared with open access cholangiographies. The Kasai procedure was performed on all patients where BA was recognized. Cholangiography was applied to 15 patients (6 laparoscopic, 9 open) during this time. The mean age of the LC patients was 47.2 days (30-75), whereas the mean age of the open cholangiography patients was 75 days (60-105). The mean age of laparoscopic cholangiographies were significantly smaller than the mean age of open cholangiographies (p=0.016). From 6 LC's, 3 patients had BA. From 9 open cholangiographies patients, 3 patients had BA. The mean time of length of stay of patients who underwent LC with no diagnosis of BA was 1.3 days (1-2 days) and they were discharged uneventfully for the further evaluation of their jaundice. However the mean time of hospital stay was 4.2 days (3-6 days) for open cholangiographies which are not BA. The mean length of hospital stay of patients who underwent LC is significantly shorter than the open cholangiographies in cases that the BA is excluded (p=0.019). In the literature there is limited data concerning LC's in children. LC is an effective alternative method in the diagnosis of BA and avoids unnecessary laparotomy. The mean length of hospital stay is after LC is considerable less than the open cases in patients which BA is excluded.

The aim of this study was to discuss our clinical experience on laparoscopic

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1. Introduction

Biliary atresia (BA) is a progressive disease that is characterized by the obstruction of the intra- and extrahepatic biliary tract (Pall and Jonas, 2005). It is the third most common cause for neonatal hepatitis and the most common cause of liver transplantation in children is a potentially lethal disease, with persisting conjugated jaundice and pale stools during the neonatal period (Balistreri, 1985). It is a rare disease, seen in 1 in about 17.000 live births (Davenport et al., 2013). The Kasai procedure, consisting of reconstruction using a Roux loop [Kasai portoenterostomy (KPE)], is performed to restore bile flow and preserve the native liver by excision of solid extrahepatic biliary remnants (Kasai et al., 1975). Prompt and accurate diagnosis of BA in a child with neonatal jaundice is very important because the timing of the Kasai procedure determines its success. Neonatologists and pediatric surgeons play a crucial role in the timely diagnosis and treatment of prolonged jaundice in newborn infants.

Until the last decade, laparotomy was necessary for accurate diagnosis. With the improvement of minimal invasive surgery in infants, laparoscopic cholangiography (LC) was introduced (Nose et al., 2005). However, there are not much studies reporting LC's in infants in literature. Therefore, we aimed to evaluate our patients with LC and compared with those of open cholangiographies. We also reviewed the literature.

2. Patients and methods

Cholangiographies for the differential diagnosis of BA between 2008 and 2013 were evaluated retrospectively. Cholangiography was applied to 15 patients (6 laparoscopic, 9 open). All patients showed an atrophic gallbladder on ultrasonography and computerized tomography (CT) scan. The diagnosis of BA was inconclusive according to conservative techniques.

Technique

After the induction of general anesthesia, an open Hasson technique was used to introduce 5mm trocar for a 30° telescope. Carbon dioxide was insufflated to a pressure of 10 to 12 mmHg. The liver status and the presence of gallbladder were evaluated. A liver biopsy was performed if needed. A 16-gauge cannula was inserted transcutaneously into the gallbladder under direct vision and contrast material (Urovist-Angiografin, Schering) was administrated. If passage of the contrast material was observed in the proximal biliary tracts and the intestinal system, the diagnosis of BA was eliminated. If no passage of the contrast material was observed, the diagnosis of BA was made and an open surgery (Kasai procedure) was performed (Senyüz et al., 2001). The duration of the procedure was 15-18 minutes. In patients with eliminated diagnosis of BA, further evaluation for prolonged jaundice was performed by gastroenterologists.

In open technique after a right subcostal incision and the exploration of the gallbladder, the top of the bladder is hitched and a vascular catheter is inserted into the fundus of the gallbladder. A small amount of bile is aspirated to check the proper position of the catheter. Then contrast is injected. The passage of contrast from the gallbladder to the intestines and intrahepatic biliary tree required to confirm the patency of the biliary tract and the diagnosis of BA is eliminated. The duration of this procedure is 30-45 minutes. Demographic data, cholangiographic diagnosis, findings at laparoscopy and laparotomy, length of hospital stay were analised using SPSS programme.

3. Results

Between 2008 and 2013, 8 boys and 7 girls with prolonged jaundice were evaluated for cholangiography (6 laparoscopic, 9 open). In patients with BA the liver was irregular and greenish-colored, while the gallbladder was fibrotic and atretic.

The mean age of the LC patients was 47.2 days (30-75), whereas the mean age of the open cholangiography patients was 75 days (60-105). The mean age of LC's were significantly younger than the mean age of open cholangiographies. (p=0.016). Total bilirubin values ranged from 7.2 to 9.1 mg/dL and direct bilirubin values ranged from 3.8 to 4.9 mg/dL.

Out of 6 LC patients, 3 patients had BA. Out of 9 open cholangiography patients, 3 patients had BA. The mean time of hospital stay of the patients who underwent LC and had no diagnosis of BA was 1.3 days (1-2 days). However the mean time of hospital stay was 4.2 days (3-6 days) for open cholangiographies in patients that the diagnosis of BA is discriminated. The mean length of hospital stay of patients who underwent LC is significantly shorter than the open cholangiographies in cases that the BA is excluded (p=0.019).

4. Discussion

BA is one of the causes of prolonged neonatal jaundice. Advanced age at the time of surgery is thought to have a detrimental effect on the post-operative prognosis (Mieli-Vergani et al., 1989; Ohi et al., 1990; Mowat, 1996; Lykavieris et al., 2005; Serinet et al., 2006). Therefore timely diagnosis of BA and differential diagnosis of neonatal jaundice have a particular importance to eliminate the delay of surgical correction which is associated with poor long-term prognosis (Mieli-Vergani et al., 1991). Methods such as radionuclide biliary tract scans (Gilmour et al., 1997) and magnetic resonance cholangiopancreatography (MRCP) (Kim et al., 2010) have poor sensitivity and specificity for BA. Recently, endoscopic retrograde cholangiography has been offered as a nonoperative method of choice for differentiating BA from other forms of cholestasis, but it is not widely used (Shteyer et al., 2012).

Laparoscopy has gained acceptance in many biliary system pathologies recently (Yamataka et al., 2012). In the last decade, laparoscopy started to be performed instead of laparotomy for the diagnosis of prolonged neonatal jaundice.

Senyüz et al. (2001) reported 17 cases with LC. They experienced no passage in 7 of 17 cases and performed the Kasai operation in those cases. In 10 cases, they were able to avoid unnecessary laparotomy (Senyuz et al., 2001). The literature contains few reports on LC. Table 1 lists the existing literature where LC has been performed for BA.

| Table 1. Literature for laparoscopic cholangiographies for biliary atresia | | | | | | |
|--|--------------|-----------------|-------|-------|-------------------------|--|
| Year | Author | Patients (n) | BA(+) | BA(-) | Right subcostal port | |
| 1983 | Costa F | 23 | 13 | 10 | - | |
| 1992 | Grigovich IN | 2 | 2 | | - | |
| 2000 | Hay | 33 | 21 | 12 | - | |
| 2001 | Senyuz OF | 17 | 10 | 7 | - | |
| 2005 | Nose S | 4 | 3 | 1 | + | |
| 2006 | Okazaki | 18 | 18 | | + | |
| 2009 | Tang ST | 38 | 30 | 8 | + | |
| 2010 | Huang L | 123 | 102 | 21 | + | |
| 2013 | Houben | 18 | 8 | 10 | - | |

In 2006, Okazaki et al. (2006) reported 18 patients in whom they performed LC, but their method differed from ours: they made a 5mm subcostal incision rather than use the umbilical port. They used this incision for visualization of the liver and the gallbladder and also exteriorized the gallbladder through this incision to inject the contrast (Okazaki et al., 2006). This method provides the direct visualisation of the gallbladder which makes the evaluation of the atresia easier and also provides the visualisation of the liver.

Tang et al. (2009) also reported the use of twoport cholangiography. They evaluated 38 patients; among them, 12 patients had good-sized gallbladders and underwent cholangiography via the exteriorized fundus. Another 8 patients had infantile hepatitis syndrome or cholestatic syndrome, 2 had BA, and 2 had biliary hypoplasia. In their series, 5 patients' gallbladders dissected from the liver bed underwent cholangiography, and BA in 3 cases and biliary hypoplasia in 2 cases were identified. The remaining 21 had atretic gallbladders and varying degrees of liver fibrosis. Cholangiography via the exteriorized fundus was abandoned and converted to open Kasai portoenterostomy. They proposed laparoscopy-assisted cholangiography as a simple, accurate, and safe method for the diagnosis of prolonged jaundice in children (Tang et al., 2009).

The largest series belongs to Huang et al., (2010) who evaluated 144 patients laparoscopically for prolonged jaundice. LC failed in 21 cases (14.6%) where atrophic gallbladder was found, and BA was confirmed by subsequent laparotomy. For the remaining 123 cases, BA was diagnosed in 88 (71.5%), biliary hypoplasia in 14 (11.4%), and cholestasis in 21 (17.1%) (Huang et al., 2010).

Recently, Houben et al. (2013) defined a simple method for LC consisting of hitching the gallbladder to the lateral abdominal wall with a prolene suture and then performing the cholangiography (Houben et al., 2013).

Conclusion

LC provides exact diagnosis with a minimally invasive procedure. It avoids unnecessary laparotomy in cases whose diagnosis is suspicious. Eventhough the mean ages of the LC's are significantly younger than the mean age of the open cholagiographies the mean length of hospital stay is after LC is considerable less than the open cases in patients which BA is excluded.

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