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**Precise mapping of hilar cholangiocarcinoma with a skip lesion by SpyGlass
Cholangioscopy - A case report**

Chiang *et al.* Pre-operative-mapping of hilar cholangiocarcinoma.

Abstract

BACKGROUND

Cholangiocarcinoma (CC) is a very aggressive cancer with poor prognosis. As surgery is the only curative therapy, preoperative evaluation of tumor extent is essential for surgical plan. Although, image modalities with high quality such as computed tomography and magnetic resonance image have been used in preoperative evaluation for longtime, the accuracy is not so high. Precise localization of tumor spread arising from hilar region before operation is still an unmet need.

CASE SUMMARY

The 52-year-old woman presented to our emergency department with jaundice, abdominal pain, and fever. Initially, she was treated as cholangitis, and endoscopic retrograde cholangiopancreatography (ERCP) with the cholangiogram showing long segment filling defect in common hepatic duct (CHD), with dilatation of bilateral IHDs. Transpapillary biopsy was performed, and the pathology showed the result of intraductal papillary neoplasm, with high-grade dysplasia. After procedure for treatment of cholangitis, contrasted-computed tomography (CT) study revealed hilar lesion with undetermined Bismuth-Corlette classification. The SpyGlass cholangioscopy showed the lesion involved the confluence of CHD with one skip lesion in posterior branch of right IHD which cannot be detected by previous image modalities. The surgical plan was changed from extended left hepatectomy to extended right hepatectomy. The final diagnosis was hilar CC, pT2aN0M0. Till now, she has been in disease-free status for more than three years.

CONCLUSION

Spyglass cholangioscopy may have a role on precision localization of hilar CC to provide surgeon more information before operation.

Key Words: Case report; Hilar cholangiocarcinoma; Jaundice; Spyglass cholangioscopy; Bismuth-Corlette classification; Hepatectomy

Chiang CH, Chen KC, Devereaux B, Chung CS, Kuo KC, Lin CC, Lin CK, Wang HP, Chen KH. Precise mapping of hilar cholangiocarcinoma with a skip lesion by SpyGlass Cholangioscopy – A case report. *World J Gastrointest Surg* 2023; In press

Core Tip: The precise localization for hilar CC is important for surgical planning. This case highlighted the important role of Spyglass cholangioscopy over this issue. However, Spyglass cholangioscopy may be difficult to insert during the first encounter because of obstruction. Two-steps approach for obstructive jaundice caused by hilar CC was proposed here: 1. Insertion of biliary plastic stents first for relieving jaundice and dilating the stricture site 2. Removal of biliary plastic stents after relieving jaundice and subsequent examination of hilar CC involvement by Spyglass cholangioscopy.

INTRODUCTION

Since the first clinical usage of SpyGlass cholangioscopy (Microvasive Endoscopy, Boston Scientific Corp, Natick, MA) reported in 2007, numerous clinical study of SpyGlass system have been published(1). However, to our best knowledge, no definite guideline was made for application of SpyGlass cholangioscopy in CC mapping. One study showed that overall procedure success rate was up to 89%, and the yielding rate of device-assisted biopsy for histological examination was up to 88%(2). Another study showed that the accuracy of SpyGlass visual impression for differentiating malignant from benign ductal lesions was 89%, and targeted biopsies accuracy rate was 82%(3).

Many patients with early stage of hilar CC, who accepted radical left or right extended hepatectomy, had high recurrent rate, up to 52% in three years after R0 resection(4). The skip lesion of intraepithelial neoplasia and intraductal papillary neoplasm in bile duct, or indeterminate classification of CC could lead the consequence of high recurrence rate(5, 6). Since the advent of SpyGlass cholangioscopy with high

resolution image of bile duct, it may provide some role on precise localization of hilar CC. The information of SpyGlass cholangioscopy may provide surgeons for more delicate surgical planning.

Herein, we reported a case of hilar CC which was precisely mapped by SpyGlass cholangioscopy with detection of one skip IHD lesion. In addition, we demonstrated the potential benefit of SpyGlass cholangioscopy in the field of precise localization of hilar CC before operation to guide surgical plan.

CASE PRESENTATION

Chief complaints

The 52 years-old woman without any underlying disease presented with acute onset of right upper quadrant abdominal pain for one day.

History of present illness

Before her presentation, progressive tea-colored urine and generalized yellowish skin with itching sensation developed for 3 days. Besides, she had fever up to 38 degrees Celsius.

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History of past illness

She denied any past illness.

Personal and family history

She denied any personal and family history.

Physical examination

Physical examination showed right upper quadrant tenderness without rebounding pain.

Laboratory examinations

The biochemistry test revealed hyperbilirubinemia (12.5 mg/dL, normal range 0.2 ~ 1.5 mg/dL).

Imaging examinations

Subsequent abdominal sonography revealed acute calculous cholecystitis, suspected common bile duct (CBD) stone with dilated CBD and bilateral IHDs (Figure 1A). For relieving her symptoms, both ultrasound-guided percutaneous gallbladder drainage and ERCP were performed. The cholangiogram revealed a long filling defect from proximal CBD to common hepatic duct (CHD) confluence (Figure 1B). Moreover, some tissue fragments were extracted out *via* balloon lithotripsy during ERCP exam, and some of them were sent for pathological examination. Besides, brushing cytology was also done. Endoscopic retrograde biliary drainage (ERBD) was placed into right side IHD for relieving her jaundice. Furthermore, subsequent contrast-enhanced computed tomography revealed similar findings as ERCP (Figure 1C). Later, left percutaneous transhepatic cholangiography drainage was performed for relieving unresolved jaundice after ERBD (Figure 1D). Since then, her hyperbilirubinemia was resolved gradually, and then final pathology of CHD lesion showed intraductal papillary neoplasm, with high-grade dysplasia.

FINAL DIAGNOSIS

Cholangiocarcinoma pT2aN0

TREATMENT

After serial work-up and management, a general surgeon was consulted for the curative surgery. Initially, extended left hepatectomy was planned for the patient, because this patient's right lobe is larger than left side, and it could preserve more liver without any pre-operative portal vein embolization.

However, surgical plan was changed after examination of SpyGlass cholangioscopy which revealed intraductal mass lesion, from CHD to the margin of bifurcation of IHDs

(Figure 2A), with one skip lesion in right posterior branch of bile duct (2 cm proximally away from bifurcation of CHD) (far and near views in Figure 2B and Figure 2C respectively). Besides, the cholangioscopy showed normal appearance of mucosa in proximal left IHD (Figure 2D). Based on these findings, the surgeon changed the initial surgical plan of extended left hepatectomy (LH) to extended right hepatectomy (RH). Before the operation, embolization of right portal vein was performed because the volume of left lobe of liver was not supposed to be adequate after extended RH. After confirmation of atrophy of right lobe of liver and enlargement of left lobe of liver four weeks later by image with acceptable indocyanine green retention ratio (15 minutes:6%, normal range < 10 %), laparoscopic extended RH was performed uneventfully eight weeks later after portal vein embolization. The surgical specimen showed one skip lesion in the right-posterior branch hepatic duct which was compatible with Spyglass cholangioscopy findings(Figure 3A, arrow). The histology showed well differentiated cholangiocarcinoma which invaded beyond the wall of the bile duct to surrounding adipose tissue (Figure 3B).

OUTCOME AND FOLLOW-UP

Till now, the patient is quite well-being, and her regular liver tri-phasic CT showed free of disease for more than three years.

DISCUSSION

CC is the second common hepatic malignancy, just after hepatocellular carcinoma (HCC). In the group of CC, hilar CC is the most common in all group of CC, approximately 50-60% of all CC(6). The only curative treatment is extended semi-hepatectomy with hilar resection. Therefore, choosing left-side or right-side extended hepatectomy is the most crucial issue for all surgeons, and it depends on precise localization of hilar lesion beforehand. Before 2007, ERCP plus tri-phase dynamic CT or magnetic resonance cholangiopancreatography (MRCP) were the only methods to localize lesion and perform pre-operative mapping(7, 8).

Since Spyglass cholangioscopy system first appeared in 2007, many studies have shown its diagnostic and therapeutic values over indeterminate biliary strictures(9-11). However, to our best knowledge, there was no study targeted on pre-operative mapping for hilar CC before. According to the previous studies and case series, high recurrent rate of hilar CC might result in skip lesion or residual intraductal papillary neoplasm of the bile duct on the other side of IHD(4). The case we demonstrated here showed one skip residual intraductal papillary neoplasm in right posterior branch of bile duct. The cholangioscopic findings provided the valuable information for surgeons to make surgical planning. In this case, for Bismuth type I or type II hilar CC, extended LH might be the rational surgical planning for operator because extended LH and extended RH with en-bloc resection demonstrate similar long-term survival and extended LH hilar en-bloc resections are more feasible and safer peri-operatively and post-operatively(12). However, because of more precise information of tumor extent provided by Spyglass cholangioscopy, the surgical plan was changed for this patient. Based on the experience from this case, two-steps approach for obstructive jaundice caused by hilar CC was proposed here: 1. Insertion of biliary plastic stents first for relieving jaundice and dilating the stricture site 2. Removal of biliary plastic stents after relieving jaundice and performing Spyglass cholangioscopy for examination of hilar CC involvement. Notably, cross-sectional images were still required for evaluation of nodal involvements and distal metastases. Besides, for those patients with periductal-infiltrating type CC, Spyglass cholangioscopy may has limited role. Thus, combination of Spyglass cholangioscopy, cross-sectional images, and cholangiogram may render more precise information of hilar CC localization and staging for providing the most suitable treatment for patients.

CONCLUSION

Spyglass cholangioscopy may have a role on the precision localization of hilar CC for surgical planning, however a larger scale study is warranted.

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SIMILARITY INDEX

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