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Left hepatic trisectionectomy for perihilar cholangiocarcinoma with a right-sided round ligament: A case report

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Abstract

BACKGROUND

A right-sided round ligament (RSRL) is a rare, congenital anomaly of the intrahepatic portal vein, with a reported frequency of 0.2%-1.2%. For patients with perihilar cholangiocarcinoma associated with an RSRL, an accurate understanding of the vascular and biliary anatomy is indispensable.

CASE SUMMARY

We report a 70-year-old male with perihilar cholangiocarcinoma associated with an RSRL. After percutaneous transhepatic embolization of the left and anterior portal branches, we conducted a left trisectionectomy of the liver with extrahepatic bile duct resection and hepaticojejunostomy. The postoperative course was uneventful, and R0 resection was achieved. When the liver volume of each section was compared between 7 patients with an RSRL and 20 patients with normal portal vein anatomy, the posterior section in RSRL patients was significantly larger than that in patients with normal portal vein anatomy (median: 457 mL vs 306 mL, $P = 0.031$). In patients with perihilar cholangiocarcinoma associated with an RSRL, left trisectionectomy has several surgical advantages: (1) The posterior branch of the portal vein often ramifies independently, and the division of the portal vein is easily conducted; (2) A relatively large amount of remnant liver can be retained; and (3) The anatomy of the posterior branch of the Glissonian pedicle is similar to that in patients with normal anatomy.

CONCLUSION

In patients with an RSRL and perihilar cholangiocarcinoma that does not involve the posterior section, left trisectionectomy may be a favorable choice.

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Core tip: A right-sided round ligament (RSRL) is a rare, congenital portal vein anomaly. We present a case of perihilar cholangiocarcinoma in a 70-year-old male with an RSRL. Additionally, we reviewed the medical records of 7 patients with an RSRL who underwent hepatobiliary and pancreatic surgery at our hospital. Left trisectionectomy may be a favorable choice for resection in patients with an RSRL and perihilar cholangiocarcinoma because: (1) The posterior branch of the portal vein often ramifies independently; (2) The volume of the posterior section is relatively large; and (3) The anatomy of the posterior Glissonian pedicle in an RSRL is not very different from that in ordinary patients.

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INTRODUCTION

A right-sided round ligament (RSRL) is a rare, congenital portal vein (PV) anomaly with a reported frequency of 0.2%-1.2%, which has been mostly described in East Asian patients^[1,2]. RSRLs have also been referred to as a “left-sided gallbladder” in several reports^[3-5]. However, it is now clear that in this anomaly, the gallbladder is located in its normal position along the main portal fissure^[5]. An RSRL arises as a result of the diminishment of the left umbilical vein in the prenatal period. Consequently, the right umbilical vein forms an umbilical portion (UP)^[1]. RSRLs are classified into three types: Ppost-i, P-bifurcation, and P-trifurcation types^[6]. In the Ppost-i type, the posterior branch of the PV (Ppost) ramifies independently from the main portal trunk. In the P-bifurcation type, the main PV ramifies to the right PV and the left PV (LPV), similar to the normal anatomy, and in the P-trifurcation type, the main PV ramifies to the left, anterior, and posterior branches.

In this report, we present a case of perihilar cholangiocarcinoma with an RSRL. Additionally, the volume of each liver section was compared among 7 patients who had an RSRL and underwent hepatobiliary or pancreatic surgery at National Cancer Center Hospital, Tokyo, Japan and 20 patients who were randomly selected with normal PV anatomy. The aim of this study was to analyze the characteristic differences in the liver volume in patients with an RSRL, and to investigate the appropriate surgical procedure for patients with an RSRL and perihilar cholangiocarcinoma.

Statistical analysis was performed by using a Mann-Whitney U test. The terminology of the liver anatomy was principally based on the Brisbane 2000 nomenclature^[7]. The volumetric evaluation of each liver section was achieved manually by tracing each liver section on contrast-enhanced computed tomography (CT) or magnetic resonance imaging and determining the area. Two examiners (Ishida T and Nara S) independently performed the volumetry, and cases with marked discrepancy were resolved by discussion. A three-dimensional (3D) image analysis software system (Synapse Vincent ver2, Fujifilm, Japan) was used to confirm the results of liver volumetry.

CASE PRESENTATION

Chief complaint

Epigastric pain.

History of present illness

A 70-year-old male visited a nearby hospital with a 1-mo history of epigastric pain.

Laboratory examinations revealed an elevation of liver enzymes and total bilirubin (T-Bil: 4.5 mg/dL). Contrast-enhanced CT revealed the wall thickening of the hilar bile duct and the bilateral dilatation of the intrahepatic bile ducts. Thus, perihilar cholangiocarcinoma was suspected, and the patient was referred to our hospital.

History of past illness

The patient received medical treatment for hypertension and gout.

Personal and family history

The patient had no particular personal or family history.

Physical examination

The patient was afebrile. An abdominal examination revealed no distention or tenderness.

Laboratory examinations

At our hospital, laboratory tests revealed that the levels of total bilirubin (T-Bil), serum aspartate transaminase, and alanine aminotransferase were all within the normal limits. However, biliary enzymes including serum alkaline phosphatase and γ -glutamyltransferase were elevated, with values of 461 IU/L (normal range, 106–322 IU/dL) and 118 IU/L (normal range, 13–64 IU/dL), respectively. The white blood cell counts were slightly elevated at $9.5 \times 10^3/\mu\text{L}$ (normal range, $3.3\text{--}8.6 \times 10^3/\mu\text{L}$), but the serum C-reactive protein level was 0.05 mg/dL (normal range, ≤ 0.14 mg/dL). The levels of carcinoembryonic antigen (CEA) and carbohydrate antigen 19-9 (CA19-9) were slightly elevated, with values of 7.7 ng/mL (normal range, 0.0–5.0 ng/mL) and 52 IU/mL (normal range, 0–37 IU/mL), respectively. The indocyanine green retention rate at 15 min after injection was 7.7% (normal range, $\leq 10\%$).

Imaging examinations

On contrast-enhanced CT images, the biliary wall was thickened at the confluence of the left and right hepatic ducts (Figure 1A and B, white arrowhead). The biliary intrahepatic bile ducts were moderately dilated. The posterior branch of the PV ramified independently (Ppost-i type, Figure 1E and F, black arrow), followed by the ramification of the anterior branch of the PV (Pant) and the LPV (Figure 1D and F, blue arrow). The Pant formed the right-sided UP (Figure 1A, B, and F, blue arrowhead) and connected to the round ligament (Figure 1C, black arrowhead). The PV branches to the anterior section diverged from the UP (Figure 1, yellow arrow). The round ligament was located on the right side of the gallbladder (Figure 1E). The 3D image of the PV clearly illustrated the independent ramification of the posterior branch (Ppost-i) and the subsequent bifurcation of the Pant and LPV (Figure 1F). Endoscopic retrograde cholangiography revealed the severe stenosis of the hilar bile duct (Figure 2, yellow arrow). The stenotic portion extended from the hepatic hilum to the right, B2, and B3 bile ducts (Figure 2, white arrowheads). According to CT volumetry, the posterior section of the liver was 501 mL (42% of the total liver volume). The biopsy of the stenotic bile ducts suggested a diagnosis of adenocarcinoma.

FINAL DIAGNOSIS

Perihilar cholangiocarcinoma (Bismuth type IIIb^[8]), T2aN0M0, Stage II^[9].

TREATMENT

Operation

The patient underwent percutaneous transhepatic PV embolization (PTPE) of the left and anterior portal branches. Twenty days after PTPE, the volume of the posterior section increased up to 49%, and left trisectionectomy (including S1) with extrahepatic bile duct resection and hepaticojunostomy was performed (Figure 3). After the sequential division of the left and right anterior hepatic arteries, the root of the LPV and Pant was ligated and divided. Hepatic parenchymal resection was conducted along the right hepatic vein, and the common trunk of the middle and the left hepatic veins was stapled and divided. Finally, the posterior branch of the bile duct (Bpost) was cut, and a specimen was removed. The frozen section analysis of the bile duct stump was negative for carcinoma (both the duodenal and hepatic sides). Biliary reconstruction was performed using the Roux-en-Y method. The operation time was 9

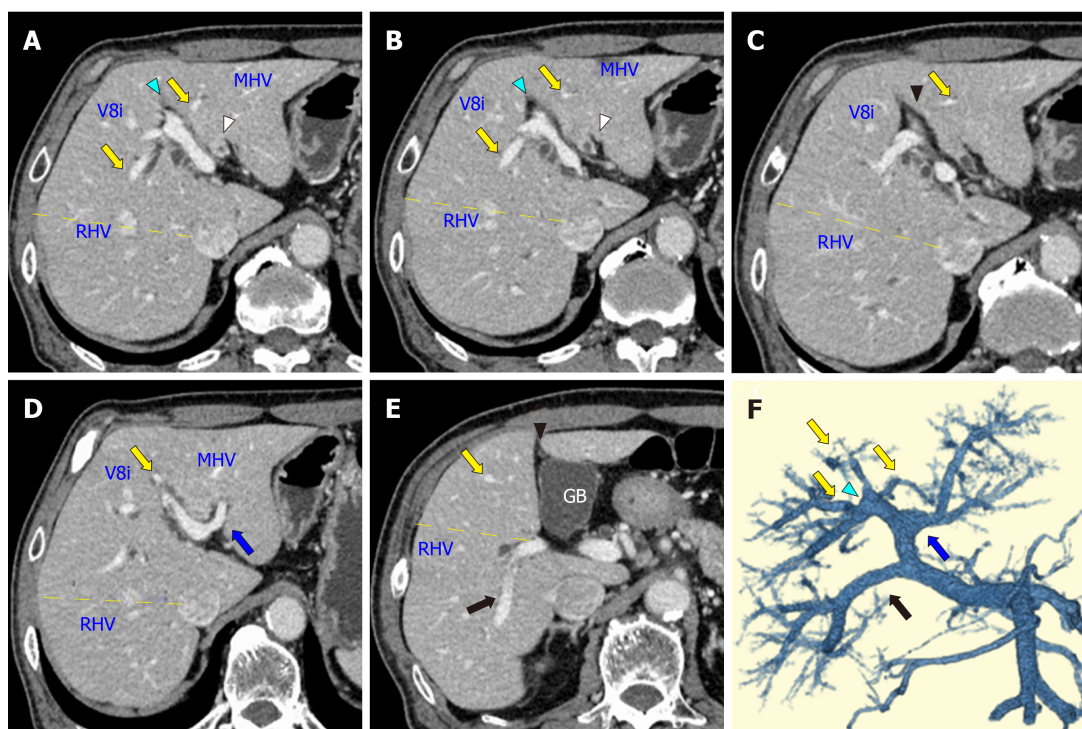


Figure 1 Computed tomography images of the patient. A: The wall of the biliary trees was thickened at the confluence of the left and right hepatic ducts (white arrowhead). The yellow dotted line indicates the boundary between the anterior and posterior sections; B: The anterior branches of the portal vein (Pant, yellow arrows) diverged from the right-sided umbilical portion (blue arrowhead); C: The round ligament (black arrowhead) was attached to the right-sided umbilical portion; D: The left portal vein (LPV, blue arrow) ramified at the hepatic hilum. The middle hepatic vein (MHV) was observed between the LPV and Pant; E: The posterior branch of the portal vein ramified independently (Ppost-i type, black arrow), and the round ligament was located on the right side of the gallbladder (black arrowhead); F: A three-dimensional image of the portal vein clearly illustrated the independent ramification of the posterior branch (Ppost-i, black arrow) from the main portal vein. RHV: Right hepatic veins; MHV: Middle hepatic vein; GB: Gallbladder.

h and 40 min, and the intraoperative blood loss was 780 mL. The postoperative course was uneventful, and the patient was discharged on the 18th day after surgery.

Pathological diagnosis

Histologically, the tumor was diagnosed as a well-differentiated adenocarcinoma extending along the perihilar bile ducts that was 4.5 cm × 3.3 cm in size. The depth of invasion was confined to the subserosal layer.

OUTCOME AND FOLLOW-UP

There was no sign of recurrence 15 mo after surgery. Carbohydrate antigen 19-9 level decreased within normal range 1 mo after surgery. However, CEA level continued to show slight elevation of 6.0-7.0 ng/mL after surgery.

Summary of 7 cases with an RSRL

The clinicopathological characteristics of 7 patients, who had an RSRL and underwent hepatobiliary or pancreatic surgery at National Cancer Center Hospital, Tokyo, Japan, are shown in Table 1, Figure 4, and Supplementary Figures 1-6. Five patients were classified as the Ppost-i type, and the other two patients were classified as the P-bifurcation type.

Comparison of liver volumetry between patients with an RSRL and normal anatomy

The volume of each liver section was compared between patients with an RSRL ($n = 7$) and normal PV anatomy ($n = 20$). CT volumetry revealed that the median volumes of the posterior section in patients with an RSRL, Ppost-i type, and P-bifurcation type were 457 mL (42%), 501 mL (44%), and 241 mL (27%), respectively. In contrast, the median volume of the posterior section in patients with normal portal anatomy was 306 mL (31%) (Table 2). The volume of the posterior section was significantly larger in patients with an RSRL of the Ppost-i type than in patients with normal PV anatomy ($P < 0.001$).

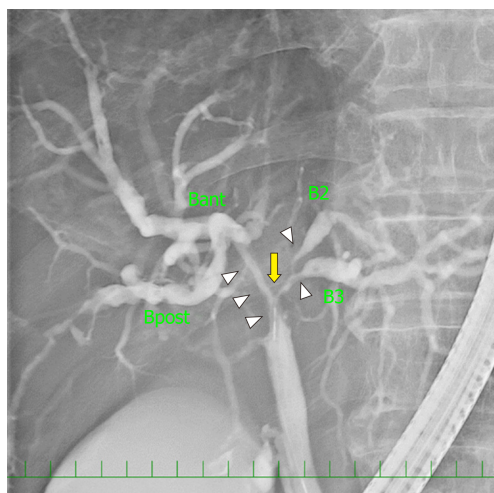


Figure 2 Endoscopic retrograde cholangiography revealed the severe stenosis of the hilar bile duct (yellow arrow). The stenotic portion extended from the hepatic hilum to the right, B2, and B3 bile ducts (white arrowheads).

DISCUSSION

During embryonic development of the liver, both right- and left-sided umbilical veins are present, but in the 8th wk of embryonic development, the umbilical vein on the right side atrophies, and the vein on the left side becomes dominant^[10]. Therefore, the UP is usually located anatomically in the left liver. Conversely, in the case of an RSRL, the left umbilical vein is believed to atrophy and the right umbilical vein is believed to persist to form a right-sided UP and round ligament. Consequently, the gallbladder is located on the left side of the round ligament. Therefore, this type of anomaly has often been referred to as a “left-sided gallbladder” in the literature.

However, in 1997, Nagai *et al*^[11] reported that in this anomaly, the location of the gallbladder is normal along the boundary of the left and right liver (Cantlie line), and it is the round ligament that is mispositioned. In RSRL cases, the careful estimation of the biliary and vascular structures is required before hepatectomy because both the biliary^[11] and portal^[12] anatomy around the hepatic hilum are different from that in normal cases. Strong *et al*^[13] reported that many patients with an RSRL did not obtain a preoperative diagnosis of an RSRL and experienced postoperative morbidity.

Among the three patterns of PV branching, the Ppost-i type is characteristic of an RSRL, with a reported incidence of 48.6%^[6]. Indeed, this pattern was the most common type in our patients [5 in our 7 patients (71%)]. The reported incidences of P-bifurcation and P-trifurcation types were 28.6% and 22.9%, respectively^[6], and each pattern was observed in 2 (29%) and 0 of our 7 patients, respectively.

In RSRL cases, the anatomy of the PV differs from normal cases. However, the constitution of the liver section/segment and venous anatomy is reported to be the same as that in normal anatomy^[14]. In summary: (1) The liver consists of four Couinaud sectors: The left lateral (S2), the left paramedian (S3+4), the right paramedian (S5+8), and the right lateral (S6+7) sectors; (2) The right, middle and left hepatic veins (RHV, MHV and LHV) run along the border of each sector; (3) The gallbladder is located along the MHV, and the gallbladder bed is along the border of the right and left liver; and (4) The RSRL separates the ventral and dorsal parts of the right paramedian sector in patients with an RSRL, similar to how the round ligament separates S3 and S4 in normal PV anatomy. Additionally, the umbilical fissure vein (UFV)^[6] and intermediate vein for S8 (V8i)^[15], both of which are located along the watersheds of the main venous trunks (*i.e.* the UFV between the tributaries of the LHV and MHV and the V8i between the tributaries of the MHV and RHV), are consistently found in patients with both an RSRL and normal PV anatomy. Indeed, the UFV and V8i were detected in all 7 of our patients.

However, the recognition of PV ramification in RSRL patients is sometimes difficult in clinical practice. Yamashita *et al*^[12] classified the configuration of the PV into two types: I-shaped and Z-shaped. In the Z-shaped type, the PV between the branching point of the Ppost and the LPV was tilted to left or almost parallel to the vertical plane. In our patients, all 5 patients with a Ppost-I RSRL were classified as having the Z-shaped type, and 2 patients with the P-bifurcation type were classified as having the I-shaped type (Table 1). Even when the UP is not markedly tilted to the right side, we should still be aware that there is a possibility of an RSRL.

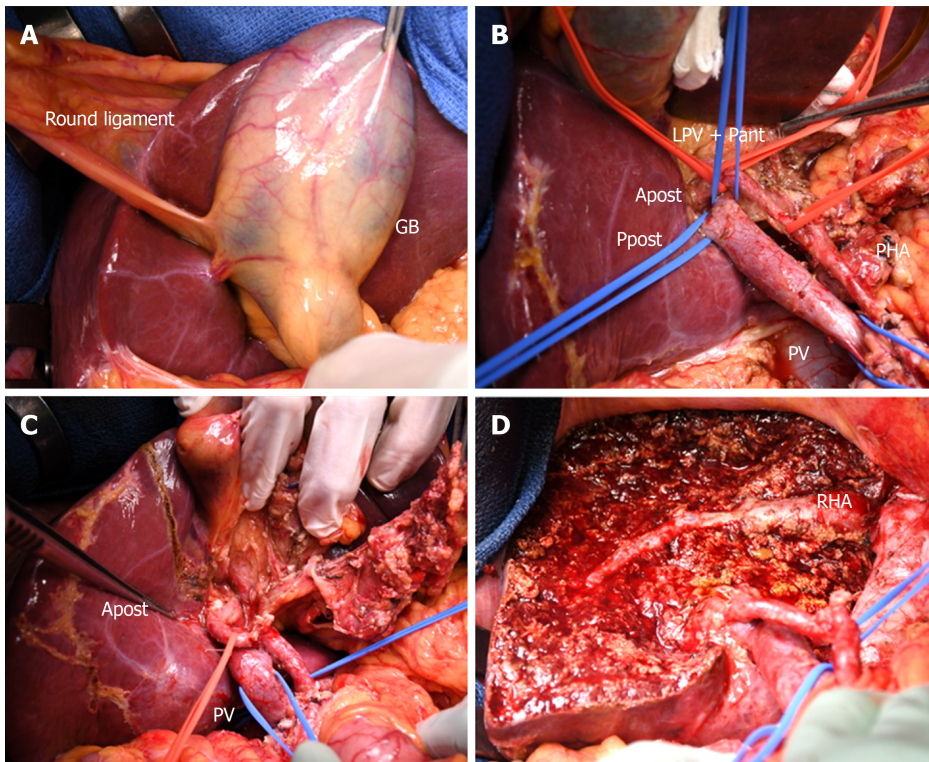


Figure 3 Operative findings. A: The gallbladder was located on the left side of the round ligament; B: The hepatic hilum was dissected, and the vessels were encircled by tape. The course of the right posterior hepatic artery was infraportal; C: After division of the left hepatic artery, right anterior hepatic artery, left portal vein and right anterior portal vein, the resection line of the liver was marked along the demarcation line on the liver surface; D: The cutting surface after left trisectionectomy and caudate lobectomy. Liver resection was conducted along the right hepatic vein. PV: Portal vein; LPV: Left portal vein; GB: Gallbladder.

With respect to the anatomy of the hepatic artery in patients with an RSRL, Shindoh *et al*^[6] noted that, in most instances, the hepatic arteries for the left hemiliver branched from the proper hepatic artery or from the left gastric artery. In our cases, 4 of 7 patients had this type of configuration. No specific correlation between PV ramification and arterial branching was identified^[6]. In addition, Nishitai *et al*^[11] reported that four biliary confluence patterns were identified in patients with an RSRL, and no correlation was observed between the biliary confluence pattern and the portal ramification pattern. In our patients, the biliary confluence anatomy was obtained in only 4 patients, and no relationship was detected between the biliary anatomy and portal ramification (Table 1). According to developmental embryology, the intrahepatic biliary ducts and arteries are formed in the stages after PV formation^[16]. Couinaud also noted that the arterial and biliary distributions did not always follow the preexisting portal ramification^[17].

In patients with an RSRL, the proportion of each liver section is reportedly different from that in the normal liver. Shindoh *et al*^[6] noted that in RSRL livers, either side of the lateral sectors [S2 and S6+7 (posterior section)] and the dorsal segment of the right paramedian sector were larger than those in patients with typical anatomy, whereas the left paramedian sector (S3+4) and the ventral segment of the right paramedian sector were significantly smaller^[6]. The reason for this observation is unknown, but because the umbilical vein is connected to the right liver, the right liver may be shifted to the left side and become enlarged, while the left liver (especially S4) may become smaller. Additionally, in a population of normal (left-sided) round ligaments, Watanabe *et al*^[18] reported that the posterior section of the Ppost-i type anatomy was significantly larger than that of the P-bifurcation anatomy ($37.4 \pm 6.1\%$ vs $27.3 \pm 5.1\%$), and 68.3% of patients with perihilar cholangiocarcinoma and Ppost-i type anatomy underwent left trisectionectomy^[18]. In our patients, the posterior section in patients with an RSRL with the Ppost-i type, but not in patients with the P-bifurcation type, was larger than that in patients with normal liver anatomy (43.9% vs 31.0%), suggesting that the Ppost-i anatomy substantially contributes to the relatively large volume of the posterior section.

Based on these findings, left trisectionectomy has several surgical advantages, especially in patients with perihilar cholangiocarcinoma associated with a Ppost-i type RSRL. First, a large liver remnant volume can be retained. Second, the ligation and division of the Pant + LPV is easy. Third, bile duct division and reconstruction

Table 1 Clinicopathological characteristics and surgical procedures of 7 patients with a right-sided round ligament at our hospital

Case	Sex	Age	Diagnosis	Operation	Portal anatomy	Portal configuration ^[1,2]	Biliary architecture ^[11]	Volumetry of the posterior/anterior/left liver
1	M	58	Hepatocellular carcinoma	Partial hepatectomy	Ppost-i	Z shape	NA	53%/32%/14%
2	M	58	Hepatocellular carcinoma	Partial hepatectomy	Ppost-i	Z shape	NA	56%/28%/15%
3	F	54	Colorectal liver metastasis	Right hemihepatectomy and choledochojejunostomy	P-bifurcation	I shape	NA	33%/30%/34%
4	M	71	Benign bile duct stenosis	Left trisectionectomy	Ppost-i	Z shape	Independent right lateral type	44%/33%/21%
5	F	41	Pancreatic head cancer	Pancreaticoduodenectomy	Ppost-i	Z shape	Total left type	41%/31%/27%
6	M	70	Perihilar cholangiocarcinoma	Left trisectionectomy	Ppost-i	Z shape	Total left type	42%/35%/21%
7	M	74	Combined hepatocellular cholangiocarcinoma	Extended posterior sectionectomy	P-bifurcation	I shape	Symmetrical type	21%/30%/44% ¹

¹In case 7, the posterior section was atrophied due to a tumor thrombus in the posterior branch of the portal vein. RSRL: Right-sided round ligament; Ppost-i: Independent branching of the posterior portal branch; P-bifurcation: Portal vein bifurcation type; NA: Not available.

can be performed in the usual manner because the biliary anatomy at the root of posterior Glissonian pedicle is distant from the complicated anatomy at the hepatic hilum associated with an RSRL, and a similar approach is possible to that of patients with a typical PV anatomy. Although left trisectionectomy is a technically demanding operation with a relatively high mortality rate^[19,20], on average, an additional 6.7 mm of bile duct can be resected compared with that in left hemihepatectomy in patients with a supraportal right posterior bile duct^[21], and left trisectionectomy has now become one of the standard procedures for perihilar cholangiocarcinoma at high volume centers^[22-24]. Therefore, left trisectionectomy may be a favorable choice for these patients, as long as there is no tumor extension to the posterior section.

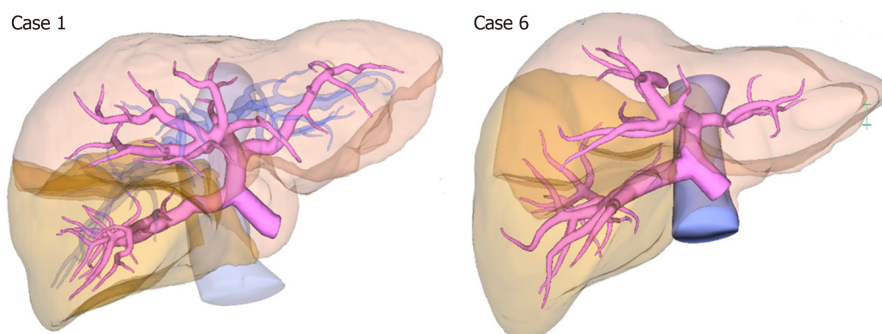
A limitation of this study was the small number of patients. To validate the results of our study, further investigations based on a large number of RSRL cases are necessary.

CONCLUSION

We presented a case of perihilar cholangiocarcinoma associated with an RSRL and reviewed 7 cases with RSRL who underwent hepatobiliary or pancreatic surgery at our hospital. The volume of the posterior section was significantly larger in patients with a Ppost-i type RSRL than that in patients with normal PV anatomy. Left trisectionectomy may be a favorable choice for patients with a Ppost-i type RSRL and perihilar cholangiocarcinoma that does not involve the posterior section.

Table 2 Comparison of the median liver volume and the percentage of each section between patients with a right-sided round ligament and normal portal vein anatomy

Liver section	RSRL, n = 7	Ppost-i, n = 5	P-bifurcation, n = 2	Normal PV anatomy, n = 20	P ¹ value	P ² value
Lateral section	247 mL (19.6%)	210 mL (17.5%)	296 mL (32.2%)	190 mL (18.5%)	0.431	0.767
Medial section	13 mL (1.0%)	12 mL (1.0%)	69 mL (6.8%)	102 mL (9.9%)	0.002	< 0.001
Anterior section	343 mL (30.8%)	371 mL (31.6%)	275 mL (29.9%)	384 mL (36.1%)	0.001	0.004
Posterior section	457 mL (41.8%)	501 mL (43.9%)	241 mL (27.2%)	306 mL (31.0%)	0.031	< 0.001
Caudate lobe (S1)	24 mL (2.3%)	23 mL (2.0%)	36 mL (3.8%)	17 mL (1.7%)	0.092	0.447
Total liver	1115 mL	1175 mL	917 mL	1032 mL	0.607	0.272

¹Right-sided round ligament *vs* normal anatomy;²Independent branching of the posterior portal branch *vs* normal anatomy. RSRL: Right-sided round ligament; Ppost-i: Independent branching of the posterior portal branch; P-bifurcation: Portal vein bifurcation type; PV: Portal vein.**Figure 4** A three-dimensional image analysis software system was used to confirm the liver volumetry. These images represent case 1 and case 6 in [Table 1](#).

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