Supplementary Information (SI).

The Impact of Pathological Cyst Wall Changes on Congenital Biliary Dilatation in Infancy

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Supplementary Figure.

Pathological findings on the luminal side of the bile duct cyst wall in a patient with iCBD (**F** in **Fig. 1**) by immunohistochemical staining (α-SMA).

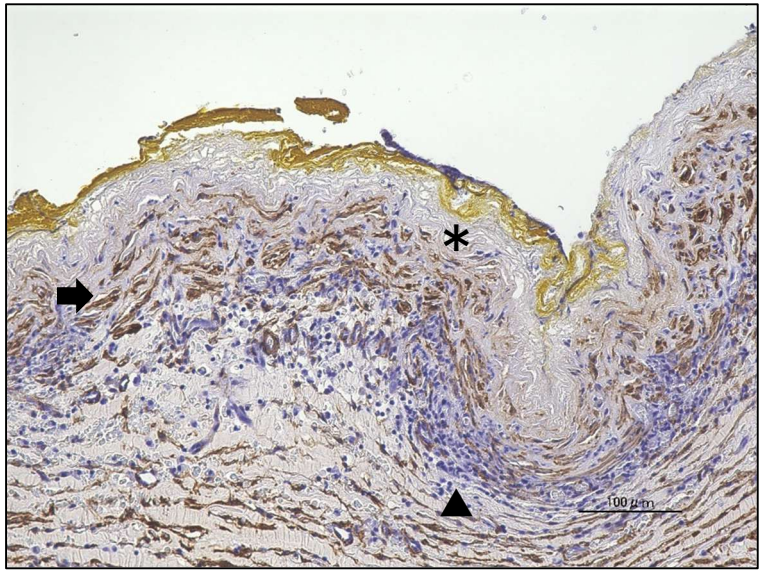
Supplementary Table.

Comparison of the clinical and pathological features among the patients with non-iCBD, iCBD without a collagen layer, and iCBD with a collagen layer.

Abbreviations: CBD, congenital biliary dilatation; iCBD, CBD patients with infiltration of inflammatory cells on the luminal side of the bile duct cyst wall; non-iCBD, CBD patients without infiltration of inflammatory cells on the luminal side of the bile duct cyst wall; IQR, interquartile range; TB, total bilirubin; DB, direct bilirubin; AST, aspartate aminotransferase; GGT, gamma glutamyl transpeptidase; TBA, total bile acid; APRi, AST to platelet ratio index.

Supplementary Figure.

Pathological findings on the luminal side of the bile duct cyst wall in a patient with iCBD (**F** in **Fig. 1**) by immunohistochemical staining (α-SMA).



Lamellar fibrotic changes, composing a collagen layer (**asterisk**), infiltration of inflammatory cells (**arrowhead**), and myofibroblasts (**arrow**) into the collagen layer, were extensively observed on the luminal side of the cyst with bile impregnation in a patient with iCBD.

Supplementary Table.

Comparison of the clinical and pathological features among the patients with non-iCBD, iCBD without a collagen layer, and iCBD with a collagen layer.

		non-iCBD (a) n=6	without collagen layer (b) in iCBD patients, n=4	with collagen layer (c) in iCBD patients, n=4	*p*-values (a) vs. (b)	*p*-values (a) vs. (c)	*p*-values (b) vs. (c)
Preoperative and intraoperative clinical findings							
Initial symptoms [Jaundice or acholic stool / Bleeding / None], n		1 / 1 / 4	3 / 1 / 0	4 / 0 / 0	0.143	***0.048***	0.686
Preoperative blood test (IQR)	TB, mg/dL	0.4 (0.3-4.4)	5.5 (2.0-10.4)	2.7 (0.9-4.4)	0.106	0.194	0.343
	DB, mg/dL	0.1 (0.1-0.4)	3.6 (1.4-6.8)	1.7 (0.5-3.0)	***0.015***	0.139	0.343
	AST, U/L	32 (27-43)	294 (83-496)	104 (69-138)	***0.010***	***0.010***	0.686
	GGT, U/L	32 (20-455)	530 (333-994)	1017 (688-1382)	0.069	0.109	0.486
	TBA, μmol/L	5 (5-9)	61 (50-80)	82 (64-119)	***0.024***	***0.010***	0.629
	APRi	0.25 (0.22-0.27)	2.65 (0.64-5.85)	0.75 (0.63-1.14)	***0.010***	***0.010***	0.686
Presence of sludge in the cyst, n (%)		2 (33)	4 (100)	4 (100)	0.076	0.076	1.000
Preoperative cyst size, mm (IQR)		31 (28-54)	45 (36-58)	60 (47-76)	0.610	0.087	0.486
Date of surgery, days (IQR)		118 (72-129)	81 (68-99)	106 (96-115)	0.352	0.610	0.343
Todani classification in type Ia / IV-A, n		4 / 2	2 / 2	2 / 2	1.000	1.000	1.000
Cyst amylase, U/L (IQR)		714 (81-1852)	3 (2-9)	3 (2-8)	***0.040***	***0.038***	1.000
Pathological findings of the cyst wall							
Presence of inflammatory cells infiltration, n (%)		0 (0)	4 (100)	4 (100)	-	-	-
Presence of collagen layer formation, n (%)		0 (0)	0 (0)	4 (100)	-	-	-
Pathological findings of the liver							
Liver fibrosis [F0 to F1 / F2 to F3], n		6 / 0	1 / 3	0 / 4	***0.048***	***0.005***	1.000
Ductular reaction [None to Mild / Moderate to Severe], n		6 / 0	2 / 2	0 / 4	0.133	***0.005***	0.429
Presence of bile plug in the intrahepatic bile duct, n (%)		1 (17)	3 (75)	3 (75)	0.190	0.190	1.000
Presence of ballooning of the hepatocytes, n (%)		1 (17)	3 (75)	1 (25)	0.190	1.000	0.486

Normal upper limits of the blood test results: TB < 1.6 mg/dL, DB < 0.4 mg/dL, AST < 31 U/L, GGT < 65 U/L, and TBA < 9 μmol/L.