



Contents lists available at ScienceDirect

## International Journal of Surgery Case Reports

journal homepage: [www.casereports.com](http://www.casereports.com)

# Laparoscopic excision of a retroperitoneal completely isolated enteric duplication cyst in an adult male: A case report and review of literature

Naoya Sasaki \*, Miru Okamura, Satoshi Kanto, Kentaro Tatsumi, Seiichi Yasuda, Atsushi Kawabe

Department of Surgery, Hikone Municipal Hospital, 1882 Hassaka-cho, Hikone-shi, Shiga Prefecture, Japan

## ARTICLE INFO

### Article history:

Received 10 February 2018

Accepted 23 March 2018

Available online 28 March 2018

### Keywords:

Completely isolated duplication cyst

Retroperitoneal duplication cyst

Laparoscopic surgery

## ABSTRACT

**INTRODUCTION:** Duplication cysts are very rare congenital malformations in adults. They are lined by gastrointestinal mucosa, connect to the digestive tract, and share smooth muscular layers and a common blood supply. In rare cases, duplication cysts are completely isolated from the digestive tract and have a proper blood supply. Completely isolated duplication cysts in the retroperitoneum are unusual so it is hard to diagnose them without a surgical resection.

**PRESENTATION OF CASE:** A 19-year-old male presented at our emergency department with sharp abdominal pain. Contrast-enhanced computed tomography detected a 5-cm multilocular cystic mass located in the retroperitoneum, caudal to the pancreatic body. The cystic mass was safely resected with laparoscopic surgery without any complication. The final pathological diagnosis was an epithelium-lined duplication cyst in the retroperitoneal space. There was no evidence of malignancy in the duplication cyst. Intracystic bleeding was assumed to be the cause of the abdominal pain.

**DISCUSSION:** The most common differential diagnoses of retroperitoneal cystic masses are pseudocysts related to pancreatitis, cysts from surrounding structures, and neoplasms. In this case, the cystic mass was diagnosed as completely isolated duplication cyst after surgical resection. It is very rarely observed in adults, but it should be listed on differential diagnoses because it has some possibility of malignancy.

**CONCLUSION:** A completely isolated duplication cyst is very rare but noteworthy because there is some possibility of malignancy, ulcerative bleeding, and perforation. A surgical resection is recommended for diagnostic treatment. Laparoscopic surgery is favorable for intraoperative inspection and it is minimally invasive.

© 2018 Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

## 1. Introduction

Duplications of digestive organs are relatively rare congenital malformations, which can arise anywhere in the gastrointestinal tract from the tongue to the anus [1–3]. It is usually detected in infancy and childhood as an acute abdomen or palpable mass, but it is rarely incidentally found in adults. Duplication cysts are characteristically lined by the gastrointestinal mucosa, and they are usually connected to the digestive tract sharing the smooth muscle layer and local blood supply [4,5]. In rare cases, the cysts are completely isolated from the digestive tract and have a proper blood supply. A completely isolated duplication cyst in the retroperi-

toneum is unusual; therefore, it is hard to diagnose until a surgical resection is performed.

Here we report the case of a young male with an isolated enteric duplication cyst in the retroperitoneum, which was safely resected via laparoscopic surgery. The cystic mass was histologically diagnosed as a completely isolated duplication cyst with internal bleeding. The clinical features, differential diagnosis, and treatment of these rare congenital malformations are also discussed.

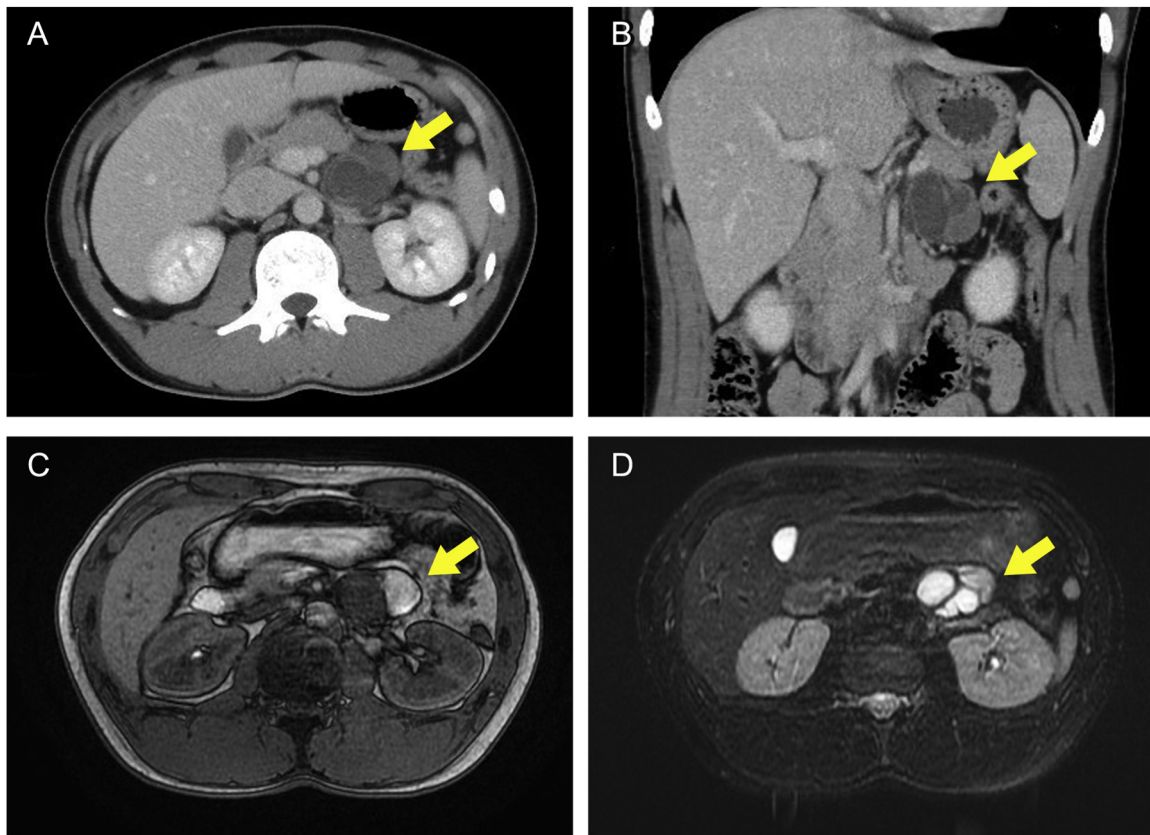
The following case report has been reported in line with the SCARE criteria [6].

## 2. Presentation of case

A 19-year-old male presented at our emergency department at midnight with a sharp pain in the upper abdomen that had persisted since that morning. There were no abnormal findings on physical examination; the abdominal wall was soft, and no

\* Corresponding author.

E-mail addresses: [naoya@kuhp.kyoto-u.ac.jp](mailto:naoya@kuhp.kyoto-u.ac.jp) (N. Sasaki), [miruokamura@gmail.com](mailto:miruokamura@gmail.com) (M. Okamura), [S.K.561001@gmail.com](mailto:S.K.561001@gmail.com) (S. Kanto), [tatsumikentaro@municipal-hp.hikone.shiga.jp](mailto:tatsumikentaro@municipal-hp.hikone.shiga.jp) (K. Tatsumi), [yasudabcd@yahoo.co.jp](mailto:yasudabcd@yahoo.co.jp) (S. Yasuda), [atsu44orca@kif.biglobe.ne.jp](mailto:atsu44orca@kif.biglobe.ne.jp) (A. Kawabe).



**Fig. 1.** Preoperative imaging detected a multilocular cystic mass in the retroperitoneum.

(A, B) Contrast-enhanced CT shows the cystic mass located in the retroperitoneum, caudal to the pancreatic body (arrows). The lateral part of the multilocular cyst had a higher CT value and internal bleeding was suspected. (C) Contrast-enhanced MRI; T1-weighted image (T1WI) and (D) T2-weighted image (T2WI). The medial part of the cystic mass shows low intensity in T1WI and high intensity in T2WI, suggesting a watery substance. On the other hand, the lateral part of the cystic mass shows higher intensity both in T1WI and T2WI, and old bleeding was suspected.

tenderness was observed. He had no remarkable past medical history. Blood cell count and serum biochemistry only showed a slight elevation in the serum total bilirubin level (1.6 mg/dL), and no inflammation or other biochemically abnormal values were detected.

Contrast-enhanced computed tomography (CT) showed a 5-cm multilocular cystic mass located in the retroperitoneum, caudal to the pancreatic body (Fig. 1A, B). A part of the cyst showed a higher CT value, which suggested internal bleeding. He was hospitalized for observation and discharged the next morning because the pain was relieved. Further blood tests showed no abnormal pancreatic enzyme values or tumor markers, including carcinoembryonic antigen, CA19-9, and DUPAN-2. Contrast-enhanced magnetic resonance imaging (MRI) also showed a multilocular cystic mass with old internal bleeding (Fig. 1C, D). The abdominal pain at the initial visit was speculated to be caused by internal bleeding of the cystic mass.

Based on these clinical examination findings, lymphangioma, teratoma, and cystic neurilemmoma were listed as differential diagnoses. Although there was no evidence of malignancy, the patient and his family strongly hoped to undergo surgical resection of the cystic mass rather than a careful follow-up. Contrast-enhanced CT taken for blood vessel evaluation 2 months after the first admission showed no significant change in the size of the cystic mass. CT angiography showed that the cystic mass had blood supply from the left renal artery (Fig. 2B) and the drainage vein flowed into the splenic vein (Fig. 2A).

A laparoscopic surgery was performed under general anesthesia. The gastrocolic ligament was incised to expose the pancreatic

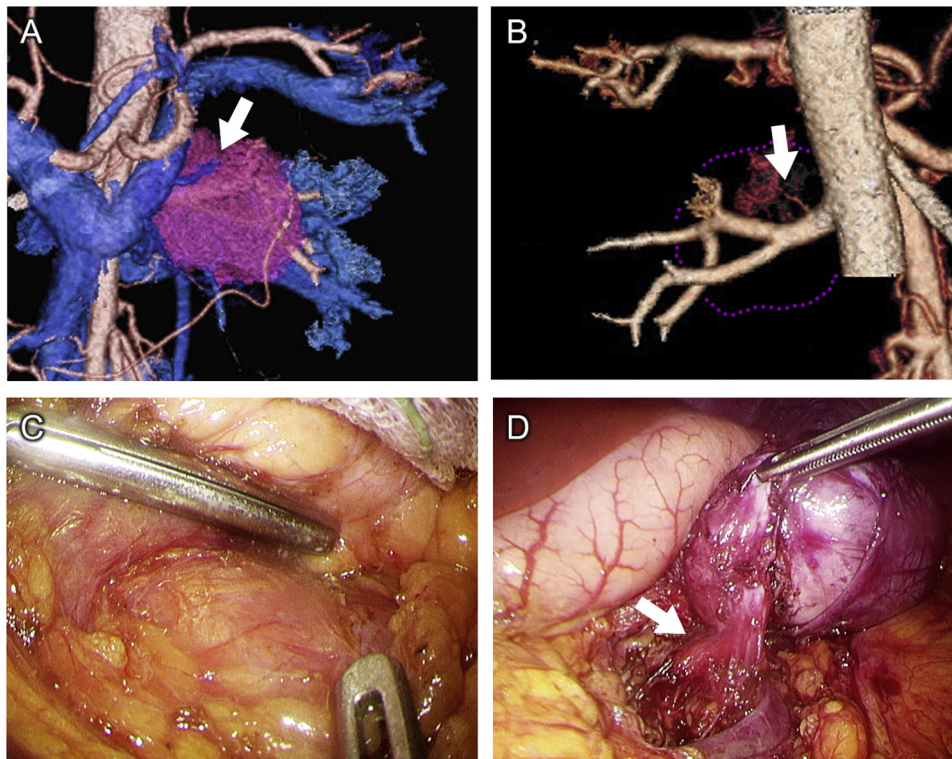
body. The retroperitoneal cystic mass was located between the pancreatic body and transverse mesocolon. The cystic mass was also confirmed by intraoperative laparoscopic ultrasonography. The cystic mass was clearly separated off from the pancreas, and the drainage vein flow into the splenic vein was clipped and cut (Fig. 2C). By dissecting the serosa of the transverse mesocolon, the cystic mass was mobilized from the retroperitoneal connective tissue, and the feeding artery from the left renal artery was clipped and cut to excise the cystic mass (Fig. 2D). The cystic mass had no connection with any part of the digestive tract. Intraoperative bleeding was minimal. The patient recovered well without any complication, and he was discharged home on postoperative day 6. The patient was healthy without any physical problems 3 months after surgery, and serum total bilirubin level had returned to normal level.

The final pathological diagnosis was an epithelium-lined duplication cyst in the retroperitoneal space. There was no evidence of malignancy. The specimen was a multilocular cyst with mucosal fluid, and one of the chambers was filled with clotted blood (Fig. 3A, B). The innermost layer was an intestine-like mucosa and showed the presence of goblet cells. A part of the lining was a flat simple columnar epithelium (Fig. 3C), and other parts showed a villous-like structure (Fig. 3D). There were two smooth muscular layers in the cystic wall and the outermost layer was surrounded by thick solid fibrous tissue.

### 3. Discussion

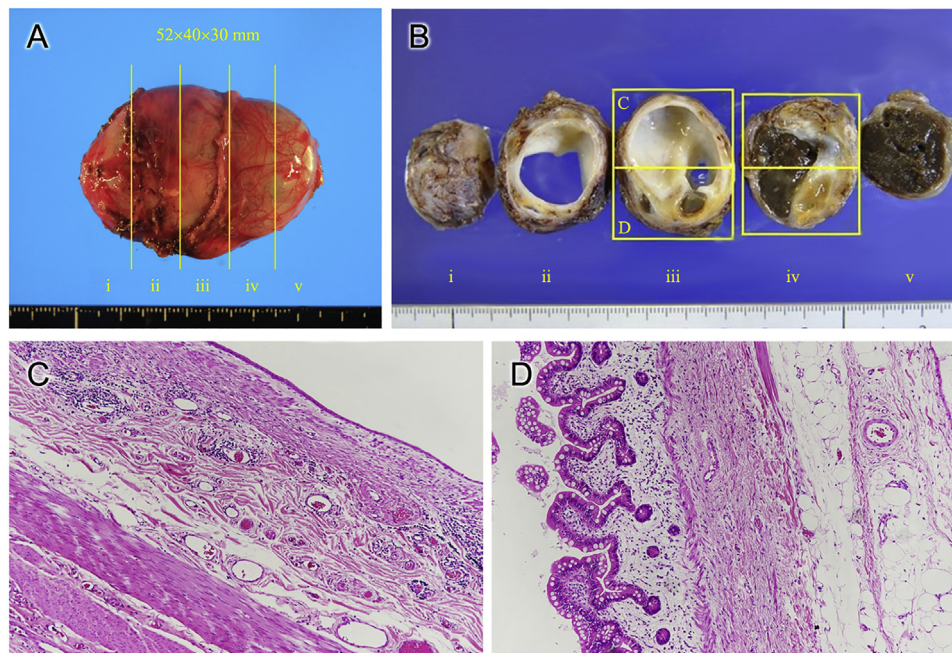
Duplications of the digestive tract are rare, 1/10,000 births, and are most commonly diagnosed at the age of 1–2 years based on





**Fig. 2.** Preoperative 3D imaging and intraoperative finding.

(A, B) A 3D reconstruction image from CT angiography; (A) The drainage vein from the upper part of the cystic mass flows into the splenic vein (arrow). (B) Dorsal view shows a feeder artery from the left renal artery runs into the back side of the cystic mass (arrow). (C, D) Laparoscopic image; (C) The spherical cystic mass was identified as caudal to the pancreatic body. (D) The feeding artery branches from the left renal artery (arrow).



**Fig. 3.** Macro- and microscopic images of the resected specimen.

(A, B) A macro image of the cystic mass; it was 52 × 40 × 30 mm multilocular cyst filled with mucinous fluid. A chamber of the multilocular cyst was filled with a blood clot. (C, D) A microscopic image; (C) Most of the cystic wall had a flat columnar epithelium. (D) Some parts of the cyst showed a villous-like structure and two smooth muscular layers. There was no evidence of malignancy.

abdominal pain or a palpable mass [1,2]. The duplications are cystic or tubular structures that can occur anywhere in the gastrointestinal tract. The most common part of the duplications is the mesenteric side of the ileum [3].

Duplications of the digestive tract typically have some connection to the gastrointestinal tract and share the blood supply with that region [4,5]. On the other hand, in a few cases, the duplication can be completely isolated from the gastrointestinal

**Table 1**

Overview of reported completely isolated duplication cysts in adults.

Age	Sex	Clinical Feature	Dimensions [cm]	Site	Surgery	Mucosal type	Reference	Year	Ref.
28	M	Incidental	not mentioned	Mesentery of the ligament of Treitz	Open	Gastric	Kim et al.	2003	[7]
64	F	Abdominal pain	7	Mesentery of the terminal ileum	Open	mucinous cystadenoma with high-grade epithelial dysplasia	Tomas et al.	2007	[8]
27	F	Abdominal fullness	9*6*1	Mesentery of the descending colon	Laparoscopic	Simple columnar epithelium	Nichols et al.	2011	[9]
28	M	Abdominal pain	25*6	Mesentery of the terminal ileum	Open		Gumus et al.	2011	[10]
20	M	Abdominal pain	3*4*2.5	Retroperitoneal, posterior to the pancreatic body	Open	Small intestine, pancreas	Emoto et al.	2011	[11]
51	M	Incidental	10*4	Mesentery of the ileum	Open	Villi, crypts, numerous mucous cells, adenocarcinoma	Blank et al.	2012	[12]
56	M	Abdominal pain	15	Mesentery of the terminal ileum	Lap. → Open	Mucinous cystadenoma, low grade	Collaud S et al.	2012	[13]
20	M	Abdominal pain and fever	7*4	Lateral region of the ascending colon	Drainage, Open	Colonic mucosa, infected	Kyriakos et al.	2013	[14]
36	F	Abdominal pain	12*8.5*6	Mesentery of the terminal ileum	Open	Columnar and gastric epithelium	Park et al.	2014	[15]
46	F	Incidental	4.5	Retroperitoneal, close to the pancreatic head	Open	Columnar epithelium	Ishige et al.	2014	[16]
52	M	Abdominal pain	4*3*3	Sub gastric	Open	Adenocarcinoma	Shin et al.	2014	[17]
48	F	Abdominal pain	6.5*4.5*2.5	Retropancreatic	Open	Jejunal	Weitman et al.	2017	[18]
19	M	Abdominal pain	52*40*30	Retroperitoneal, caudal to the pancreatic body	Laparoscopic	Small intestine	Present case	2017	

tract and have a proper blood supply [7–18]. In this case, the completely isolated duplication cyst was found in a 19-year-old male with acute abdominal pain. The cystic mass was detected with contrast-enhanced CT in the retroperitoneal space. Based on detailed examinations that included serum biochemistry and contrast-enhanced MRI, the abdominal pain was assumed to be caused by intracystic bleeding of the retroperitoneal cystic mass.

The most common differential diagnoses of retroperitoneal cystic masses are pseudocysts related to pancreatitis, cysts from surrounding structures such as mesenteric, omental, splenic, and enteric duplication cysts. Neoplasms, such as cystadenomas, mesotheliomas, and cystic degeneration from solid neoplasms, should be considered. Other non-neoplastic cysts include hematomas, urinomas, and lymphoceles [19].

In the present case, pancreatic pseudocysts and malignancy were less likely because of normal pancreatic enzyme values and the young age of the patient. However, the cystic mass was symptomatic, and surgical resection and histological analysis were the only procedures for making a diagnosis. A laparoscopic surgery was performed after cautious discussion. The cystic mass had a thick solid fibrous capsule, which was safely held with laparoscopic forceps. Proper blood vessels were identified as indicated on by preoperative CT angiography, and they were safely clipped and cut.

Completely isolated duplication cysts diagnosed in adults are extremely rare, and they have been characterized in only 13 case reports, including this case (Table 1). Most of them had symptoms of abdominal pain and three were found incidentally. Two cases diagnosed as adenocarcinomas, and one was a mucinous cystadenoma with high-grade epithelial dysplasia [8,12,17]. These completely isolated duplication cysts had gastrointestinal mucosa, two smooth muscular layers, and were surrounded by fibrous tissue. One case contained the pancreatic tissue in the solid region of the multilocular cyst, which might cause chronic pancreatitis [11]. In this case, the multilocular cyst showed internal bleeding, which is what might have caused the abdominal pain and hyperbilirubinemia, and they completely disappeared after the surgical resection.

Several theories have been proposed for the development of enteric duplication cysts. Aberrant luminal recanalization, persistent embryologic diverticula, sequestration of a part of fetal gut seem reasonable but the true etiology is open to question [20]. The mechanisms of abdominal pain are assumed to be elevated intracystic pressure due to secretions from mucosal cells, ulcers of the gastric mucosa, penetration, and pancreatitis that was caused by the aberrant pancreatic tissue in the cyst [11].

Completely isolated duplication cysts in adults are very rare; therefore, a clear indication of surgical resection is not established. However, the possibility of a malignancy based on three out of 13 case reports is not negligible. Further, ulcers may cause penetration and perforation, which can be life-threatening. Taken together, we recommend a surgical resection for two reasons: curative treatment and pathological diagnosis. Although open laparotomies were performed in many of these case reports, typical duplication cysts can be safely resected by laparoscopic surgery without any special technique and devices. Preoperative imaging modalities are indispensable; contrast-enhanced CT and MRI, CT angiography, and intraoperative laparoscopic ultrasonography are very helpful for diagnosing and identifying proper blood vessels of the duplication cyst.

#### 4. Conclusion

A completely isolated duplication cyst should be listed as a differential diagnosis of a retroperitoneal cystic mass. Surgical resection is recommended because of the possibility of malignancy. Laparoscopic surgery is preferable because the procedure is less invasive and is not complicated for well-trained general surgeons.

#### Conflicts of interest

The authors declare no conflicts of interest associated with this manuscript.

## Sources of funding

The authors do not have any funding.

## Ethical approval

Ethical approval has been exempted by our institution as publication deals with a single treated case and not a randomized trial or a case series, provided that the patient and his guardian gave their written consent both for operation and publication of the case.

## Consent

Written informed consent for the publication was obtained from the patient and his parent.

## Author contribution

NS is the first authors of this manuscript and the corresponding author. NS, SK, SY, and AK performed the surgery. KT and MO participated in perioperative management of the patient. All authors read and approved the final manuscript.

## Guarantor

Naoya SASAKI.

## References

- [1] T.R. Prasad, C.E. Tan, Duodenal duplication cyst communicating with an aberrant pancreatic duct, *Pediatr. Surg. Int.* 21 (April (4)) (2005) 320–322.
- [2] K.L. Jackson, W.J. Peche, M.D. Rollins, An unusual presentation of a rectal duplication cyst, *Int. J. Surg. Case Rep.* 3 (April (7)) (2012) 314–315.
- [3] S.R. Segal, N.H. Sherman, H.K. Rosenberg, C.L. Kirby, P.A. Caro, R.D. Bellah, et al., Ultrasonographic features of gastrointestinal duplications, *J. Ultrasound Med.* 13 (November (11)) (1994) 863–870.
- [4] R.I. Macpherson, Gastrointestinal tract duplications: clinical, pathologic, etiologic, and radiologic considerations, *Radiographics* 13 (September (5)) (1993) 1063–1080.
- [5] A. Sinha, S. Ojha, Y.K. Sarin, Completely isolated, noncontiguous duplication cyst, *Eur. J. Pediatr. Surg.* 16 (April (2)) (2006) 127–129.
- [6] R.A. Agha, A.J. Fowler, A. Saetta, I. Barai, S. Rajmohan, D.P. Orgill, for the SCARE Group, The SCARE statement: consensus-based surgical case report guidelines, *Int. J. Surg.* (Oct 34) (2016) 180–186.
- [7] S.K. Kim, H.K. Lim, S.J. Lee, C.K. Park, Completely isolated enteric duplication cyst: case report, *Abdom. Imaging* 28 (January–February (1)) (2003) 12–14.
- [8] D. Tomas, M. Zovak, S. Cicek, P. Sulentic, Z. Jukic, B. Kruslin, Mucinous cystadenoma arising in an isolated ileal duplication cyst, *J. Gastrointest. Cancer* 38 (2–4) (2007) 127–130.
- [9] K.C. Nichols, T. Pollema, M. Moncure, Laparoscopically excised completely isolated enteric duplication cyst in adult female: a case report, *Surg. Laparosc. Endosc. Percutan. Tech.* 21 (August (4)) (2011) e173–5.
- [10] M. Gümüş, M. Kapan, H. Gumus, A. Onder, S. Girgin, Unusual noncommunicating isolated enteric duplication cyst in adults, *Gastroenterol. Res. Pract.* 2011 (March (2011)) (2011), 323919.
- [11] K. Emoto, M. Mukai, S. Kawachi, Y. Kitagawa, M. Sakamoto, Duplication of the digestive organs in the retroperitoneum: a case report with reference to the importance of a standardized nomenclature and definition, *Pathol. Int.* 61 (July (7)) (2011) 430–434.
- [12] G. Blank, A. Konigsrainer, B. Sipos, R. Ladurner, Adenocarcinoma arising in a cystic duplication of the small bowel: case report and review of literature, *World J. Surg. Oncol.* 10 (April (10)) (2012) 55.
- [13] S. Collaud, C. Bayerl, G. Wille, A. Zehnder, F. Grieder, S. Meili, et al., Mucinous cystadenoma arising in a completely isolated infected ileal duplication cyst, *S. Afr. J. Surg.* 50 (March (2)) (2012) 45–46.
- [14] N. Kyriakos, C. Andreas, S. Elena, A. Charalampos, G. Chrisanthos, Infected completely isolated enteric duplication cyst management with percutaneous drainage and surgical excision after retreat of infection: a case report, *Case Rep. Surg.* 2013 (2013), 108126.
- [15] J.Y. Park, K.H. Her, B.S. Kim, Y.H. Maeng, A completely isolated intestinal duplication cyst mimicking ovarian cyst torsion in an adult, *World J. Gastroenterol.* 20 (January (2)) (2014) 603–606.
- [16] F. Ishige, O. Kainuma, H. Yamamoto, N. Takiguchi, Y. Nabeya, A. Ikeda, et al., Retroperitoneal enteric duplication cyst in an adult, *Jpn. Soc. Gastroenterol. Surg.* 47 (8) (2014) 6.
- [17] S.Y. Shin, M.Y. Cho, H. Ryu, J.W. Kim, H.S. Kim, J.M. Kim, et al., Adenocarcinoma originating from a completely isolated duplication cyst of the mesentery in an adult, *Intest. Res.* 12 (October (4)) (2014) 328–332.
- [18] E. Weitman, S. Al Diffalha, B. Centeno, P. Hodul, An isolated intestinal duplication cyst masquerading as a mucinous cystic neoplasm of the pancreas: a case report and review of the literature, *Int. J. Surg. Case Rep.* 24 (August (39)) (2017) 208–211.
- [19] J. Ahn, M.D. Chandrasegaram, K. Alsaleh, B.L. Woodham, A. Teo, A. Das, et al., Large retroperitoneal isolated fibrous cyst in absence of preceding trauma or acute pancreatitis, *BMC Surg.* 15 (March) (2015) 25.
- [20] R.L. Teele, C.I. Henschke, D. Tapper, The radiographic and ultrasonographic evaluation of enteric duplication cysts, *Pediatr. Radiol.* 10 (September (1)) (1980) 9–14.

## Open Access

This article is published Open Access at [sciencedirect.com](https://www.sciencedirect.com). It is distributed under the [IJSCR Supplemental terms and conditions](#), which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.