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CASE REPORT

Adenocarcinoma originating from a colonic duplication cyst: A case report

Jeehye Lee, Jung Wook Suh

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Jeehye Lee, Department of Surgery, Yongin Severance Hospital, Yongin 16995, Gyeonggi-do, South Korea

Jung Wook Suh, Department of Surgery, Dankook University Hospital, Cheonan 31116, Chungcheongnam-do, South Korea

Corresponding author: Jung Wook Suh, MD, PhD, Assistant Professor, Department of Surgery, Dankook University Hospital, 119 Dandae-ro, Dongnam-gu, Cheonan 31116, Chungcheongnam-do, South Korea. jickgack@gmail.com

Abstract

BACKGROUND

Gastrointestinal duplication is a rare congenital anomaly of the digestive tract, with colonic manifestations being particularly uncommon. Malignant transformation of colonic duplication cysts is rare, with adenocarcinoma being the most frequently reported type. Herein, we report a rare case of adenocarcinoma originating from a colonic duplication cyst.

CASE SUMMARY

A 49-year-old woman was found to have an elevated cancer antigen 19-9 level during a routine checkup. Imaging revealed a well-defined abdominal cavity cystic mass, which was initially suspected to be an ovarian teratoma. Laparoscopic surgery revealed a duplication cyst, and pathological examination confirmed adenocarcinoma arising from the cyst. The mass within the transverse mesocolon was successfully excised by a colorectal surgeon. Immunohistochemical analysis confirmed adenocarcinoma with invasion into the muscularis propria. Postoperative endoscopy and positron emission computed tomography scan showed no signs of malignancy, except for an elevated cancer antigen 19-9 level. A multidisciplinary team recommended no further chemotherapy, advising routine follow-up for monitoring.

CONCLUSION

Colonic duplications, though rare, remain a differential diagnosis of unexplained abdominal masses, with complete resection being their primary treatment approach.

Key Words: Gastrointestinal duplication; Colonic duplication; Surgical resection; Adenocarcinoma; Malignant transformation; Case report

Core Tip: In cases of gastrointestinal duplication involving cystic masses, maintaining a high suspicion for potential malignancy is crucial, even in asymptomatic patients. Due to the rarity of these conditions, a multidisciplinary approach is essential for accurate diagnosis and treatment. Comprehensive imaging studies, like contrast computed tomography, should evaluate the cyst's characteristics and its relation to adjacent structures. Surgical intervention, preferably through *en bloc* resection, is recommended to ensure complete removal of any neoplastic tissue. Regular follow-up and monitoring of tumor markers are vital for early detection of recurrence or progression.

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INTRODUCTION

Gastrointestinal duplication is an uncommon congenital anomaly, with gastrointestinal cystic or tubular abnormalities observed in any part of the gastrointestinal tract. The prevalence of congenital disease ranges from 1 in 4500 to 1 in 10000, with 80% of the cases diagnosed either before birth or within the first two years of life[1-4]. However, this condition may remain undetected until later in life.

The occurrence of gastrointestinal duplication has been reported in the ileum, ileocecal valve, and jejunum at rates of 30%, 30%, and 8%, respectively. However, its occurrence is rare in the colon and rectum, with incidences of 6%-7% and 5%, respectively[4-6]. If no related abnormalities are present, symptoms may not be observed until complications occur[5, 7]. Clinical symptom development depends on factors such as location, size, abnormal mucosa presence within the duplication site, adjacent bowel connection, and inflammation.

The main symptoms are abdominal pain and intestinal obstruction, while the other symptoms include the presence of an abdominal mass, chronic pain, constipation, and, less frequently, acute abdominal symptoms, including intestinal volvulus[8], intussusception, perforation, or active bleeding[9].

Herein, we present a case of adenocarcinoma arising from an asymptomatic duplication cyst in the colon of a 49-year-old female patient.

CASE PRESENTATION

Chief complaints

Detection of abnormal cancer markers.

History of present illness

A 49-year-old woman with no previous medical history presented to the gynecology outpatient clinic of a tertiary hospital with abnormal cancer markers detected during an examination at a local clinic.

History of past illness

There was no specific medical history such as diabetes, hypertension, or surgery.

Personal and family history

There was no specific personal and family history.

Physical examination

There were no abnormal findings in the physical examination upon admission.

Laboratory examinations

Elevated levels of cancer antigen (CA)-19-9: 61.6 U/mL, normal range: < 37 U/mL; CA-125: 47.3 U/mL, normal range: < 55 U/mL and normal levels of carcinoembryonic antigen: 1.1 U/mL, normal range: < 5 U/mL.

Imaging examinations

Abdominal-pelvic computed tomography (CT) revealed a well-defined 6.5 cm × 7.8 cm × 8.5 cm cystic mass with internal fatty attenuation and fluid-fluid level in the right adnexa without enhancement (Figure 1). The gynecology department at our hospital uses diagnostic laparoscopy to differentiate extragonadal teratomas, ovarian teratomas, and myxoid

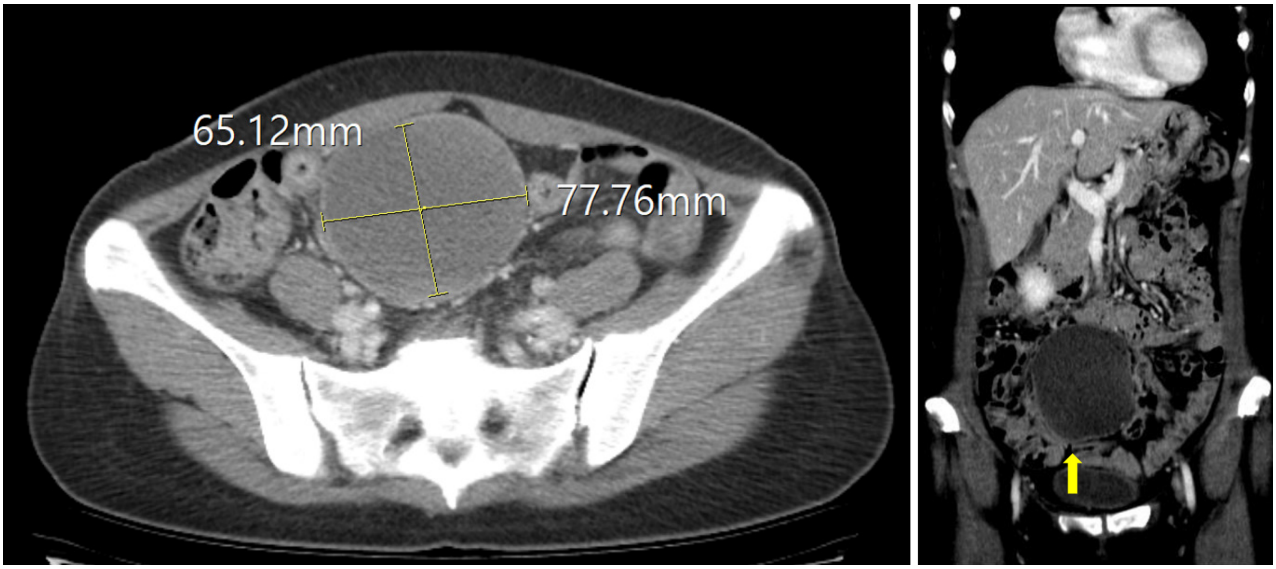

Figure 1 Preoperative radiologic findings. A 6.5 cm × 7.8 cm × 8.5 cm well-defined cystic mass with internal fatty attenuation and a fluid-fluid level was identified in the right adnexa, without enhancement. The differential diagnoses includes extragonadal teratoma, ovarian teratoma, and, less likely, myxoid liposarcoma. The absence of clear continuity with the ovary suggests an extragonadal teratoma, although an ovarian teratoma cannot be excluded if continuity is obscured by the scan slice thickness.

liposarcomas. During laparoscopy, the cyst had no communication with the ovaries; however, it was in the transverse mesocolon (Figure 2A).

MULTIDISCIPLINARY EXPERT CONSULTATION

As this case was rare, we discussed the treatment plan for the patient at a multidisciplinary treatment, consisting of a colorectal surgeon, oncologist, pathologist, radiologist, and gastroenterologist, conference and concluded that no further chemotherapy was needed due to the complete surgical resection. Regular checkups were recommended.

FINAL DIAGNOSIS

The cyst was diagnosed as adenocarcinoma originating from a colonic duplication cyst with muscularis propria invasion (Figure 3).

TREATMENT

As the tumor was hard and did not adhere to the floor and was capable of movement, a mini-laparotomy procedure was performed (Figure 2B). The cyst was then separated from the colon, mobilized, and excised by a colorectal surgeon. The well-defined cystic mass did not communicate with the colon, and no damage to the colon was observed after the resection. The patient was discharged without any postoperative complications.

Immunohistochemical analysis revealed that the tumor cells were positive for cytokeratin (CK) 7, CK 20, and caudal-type homeobox transcription factor 2 (Table 1).

OUTCOME AND FOLLOW-UP

Postoperatively, endoscopy revealed no abnormal lesions. Moreover, whole-body positron emission tomography revealed no abnormal hypermetabolic lesions suggesting malignancy (Figure 4). CA 19-9 levels measured 1 month post-surgery were 9.3 U/mL, which was within the normal range (< 37 U/mL); no other specific findings were observed. No evidence of recurrence has been observed two years post-surgery.

Table 1 Pathology results

Variables	Results
Preoperative treatment	Not done
Gross type	Other (cystic)
Tumor location	Transverse colon, mesentery
Histologic variant (World Health Organization 5th ed.)	Not otherwise specified
Histology grade	Moderately differentiated
Tumor size	9.2 cm × 8.0 cm × 7.8 cm
Invasion margins	Muscularis propria
Resection margins	Safety margin: Radial 02 cm
Lymph node	Not submitted
Lymphatic invasion	Not identified
Venous invasion	Not identified
Perineural invasion	Not identified
Tumor deposits	Not identified
Tumor budding	Negative (× 1/200)
Tumor border	Infiltrative
Stromal reaction	Absent
Preexisting adenoma	Not identified
Associated findings	None
Caudal-type homeobox transcription factor 2	Positive
Cytokeratin 20	Positive
Cytokeratin	Positive
Estrogen receptor	Negative
Paired box gene 8	Negative

DISCUSSION

Gastrointestinal duplications are uncommon congenital abnormalities diagnosed during the prenatal period or within the first two years of life in approximately 80% of cases[2,3]. Colonic duplications account for only 13% of all duplications and arise in the cecum[3,10]. Although malignant transformation in a duplication is rare, cases of adenocarcinoma, squamous cell carcinoma, carcinoid tumors, gastrointestinal stromal tumors, and leiomyosarcoma have been reported. In these rare cases, adenocarcinoma is the predominant histological malignancy type[11-14]. The duplication diagnosis is based on well-established morphological criteria, including: (1) Direct attachment to the alimentary tract; (2) Presence of a smooth muscle layer; and (3) A lining epithelium similar to that of the alimentary tract[11]. Our case met criteria 2 and 3, leading to a diagnosis of adenocarcinoma arising from a colonic duplication cyst.

The diagnosis of colonic duplications may be challenging. In children before the age of 2 years, it is often diagnosed by intrauterine ultrasonography; however, in adults, it is often revealed during surgery[3,15-17]. Contrast CT is the most effective method for establishing a diagnosis and determining the cyst composition and its interaction with adjacent structures[18]. The cysts usually have attenuation levels comparable to that of water with a discernible calcification rim in some cases[11]. The differential diagnoses include colonic duplication cyst, primary colonic malignancy, atypical gastrointestinal stromal tumor, intestinal lymphoma, myxoid liposarcoma, or loculated perforation. In women, ovarian teratoma and extragonadal teratoma should also be considered. However, a definitive diagnosis is often challenging to establish preoperatively, and surgical resection is frequently required to confirm the final pathological diagnosis[19]. In this case, a well-defined cystic mass with internal fatty attenuation and fluid-fluid level was observed; however, ovarian teratoma was more likely than colon duplication. Furthermore, nuclear medicine scans can reveal duplications in the stomach mucosa, whereas contrast enema or colonoscopy may be useful in cases in which there is a link to the colonic lumen.

Previous studies have reported colonic duplication causing intussusception, rectal hemorrhage, and pancreatitis, with abdominal pain and a palpable mass being the most common symptoms in adenocarcinoma cases[11]. In this case, the condition was detected during a regular checkup without specific symptoms.

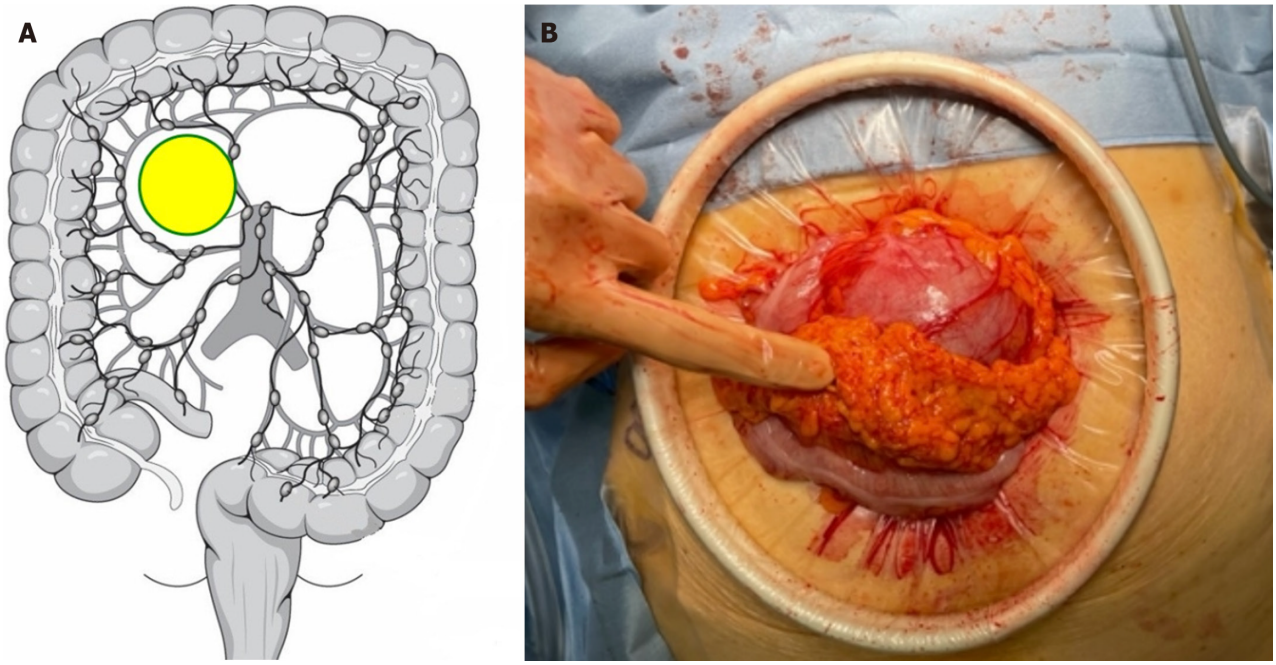

Figure 2 Colonic duplication location. A: Intraoperative findings of colonic duplication; B: Following mini-laparotomy, colonic duplication was observed adjacent to the colon and attached to the omentum.

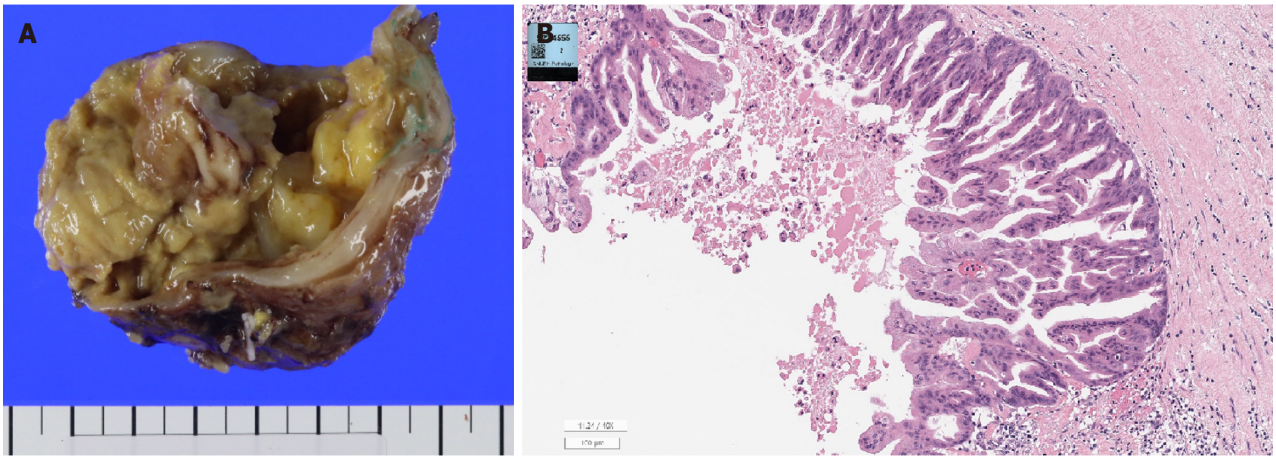

Figure 3 Gross pathologic finding. A: A cystic tumor section encased in a serosal layer revealed an internal cavity filled with mucinous-like material. Histological and pathological findings; B: The cyst wall was lined with epithelium resembling the intestinal mucosa, with mucin occupying the lumen, and mild inflammatory cell infiltration was observed in the surrounding stroma.

Colonic duplication is primarily managed through *en bloc* resection of the adjacent large intestine[2,10]. In selected cases without damage to the bowel wall or vascular supply, enucleation of duplication cysts may be performed alternatively[2]. Furthermore, advances in laparoscopic procedures and instrumentation have enabled laparoscopic resection [10]. In this case, a gynecologist attempted to approach the tumor laparoscopically and confirmed that it was distant from the uterus and ovaries and adjacent to the transverse colon. A colorectal surgeon then completed the duplication cyst resection, separating the colon and duplication cyst *via* mini-laparotomy, without damaging the colonic wall or blood vessels. Lymph node dissection was not performed because no malignancy was suspected.

The potential for colonic duplication cysts and malignancy warrants consideration of the extent of resection *via* a multidisciplinary approach; however, this team was not assembled before surgery. Following the postoperative adenocarcinoma confirmation, a multidisciplinary group was organized to assess the pathological findings and postoperative evaluations. Although lymph node dissection was not performed, further assessment revealed no increase in lymph node uptake or enlargement, and no high-risk characteristics were revealed by pathology. Therefore, complete surgical resection was considered appropriate, adjuvant chemotherapy was not administered, and routine surveillance was performed.

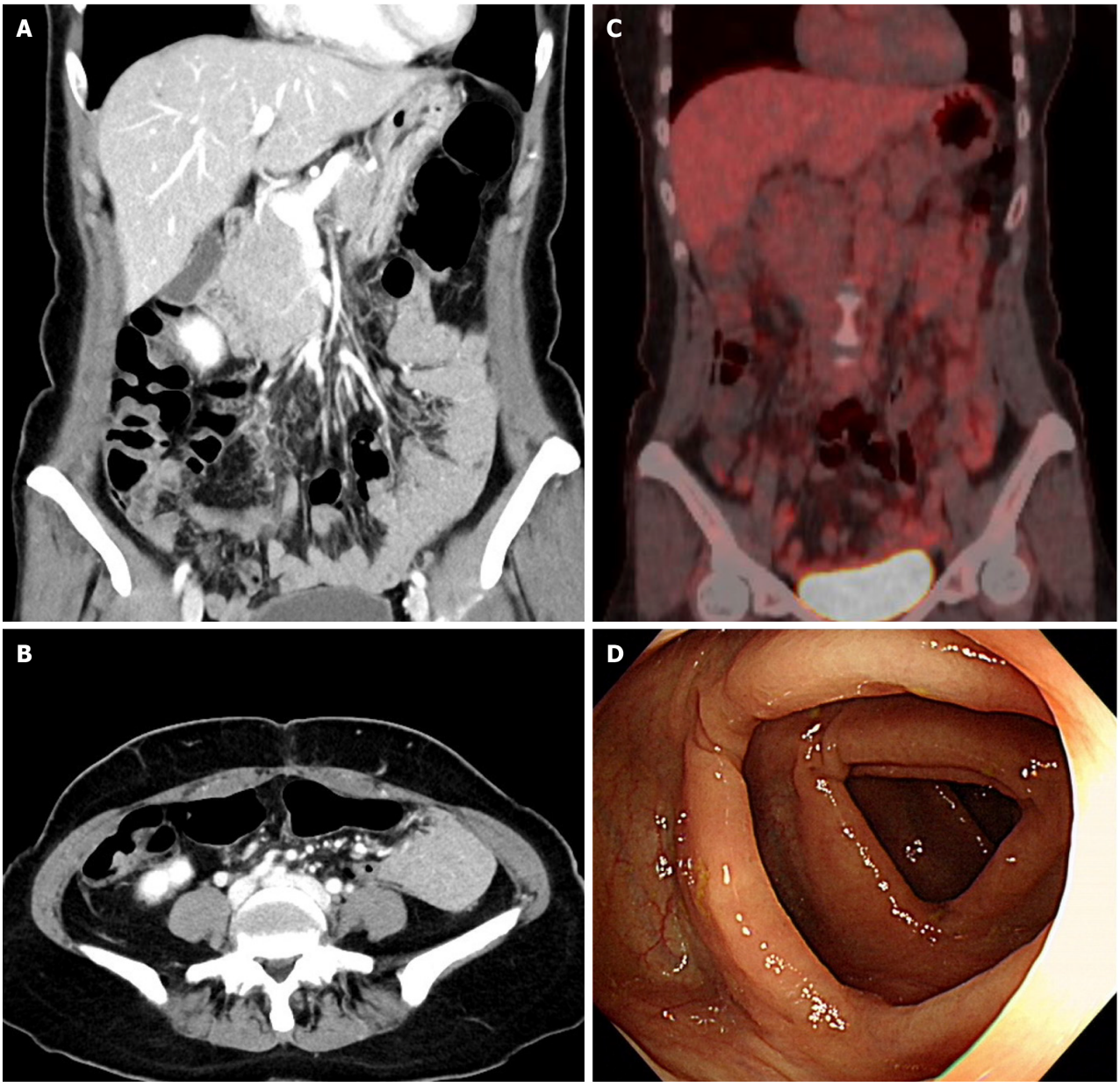

Figure 4 Postoperative imaging and endoscopy findings. A and B: Postoperative abdominal-pelvic computed tomography scans show no detectable intra-abdominal masses; C: Postoperative positron emission computed tomography-computed tomography reveals no abnormal hypermetabolic lesions suggestive of malignancy; D: Postoperative colonoscopy demonstrates no abnormal lesions in the colon.

CONCLUSION

Colonic duplication is an uncommon congenital anomaly, and malignant transformation is extremely rare. When assessing a cystic mass adjacent to or enclosed by the gastrointestinal tract, duplication should be considered in the differential diagnosis. The potential for malignancy arising from a duplication cyst should be considered, and thorough *en bloc* resection of the tumor is imperative.

FOOTNOTES

Author contributions: Lee J contributed to data curation and drafting of the manuscript; Suh JW conducted the review and editing of the manuscript.

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Country of origin: South Korea

ORCID number: Jeehye Lee 0000-0002-8050-9661; Jung Wook Suh 0000-0002-9330-9917.

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REFERENCES

1 **Schalamon J**, Schleef J, Höllwarth ME. Experience with gastro-intestinal duplications in childhood. *Langenbecks Arch Surg* 2000; **385**: 402-405 [PMID: 11127525 DOI: 10.1007/s004230000170]
2 **Ricciardolo AA**, Iaquinta T, Tarantini A, Sforza N, Mosca D, Serra F, Cabry F, Gelmini R. A rare case of acute abdomen in the adult: The intestinal duplication cyst. case report and review of the literature. *Ann Med Surg (Lond)* 2019; **40**: 18-21 [PMID: 30962926 DOI: 10.1016/j.amsu.2019.03.002]
3 **Rodríguez García P**, Sánchez Pérez A, Romera Barba E, Calero García P, González-Costea Martínez R. Colonic duplication cyst in adult. *Gastroenterol Hepatol* 2020; **43**: 360-361 [PMID: 32593469 DOI: 10.1016/j.gastrohep.2019.11.016]
4 **Kekez T**, Augustin G, Hrstic I, Smud D, Majerovic M, Jelincic Z, Kinda E. Colonic duplication in an adult who presented with chronic constipation attributed to hypothyroidism. *World J Gastroenterol* 2008; **14**: 644-646 [PMID: 18203304 DOI: 10.3748/wjg.14.644]
5 **Banchini F**, Delfanti R, Begnini E, Tripodi MC, Capelli P. Duplication of the transverse colon in an adult: case report and review. *World J Gastroenterol* 2013; **19**: 586-589 [PMID: 23382641 DOI: 10.3748/wjg.v19.i4.586]
6 **Puligandla PS**, Nguyen LT, St-Vil D, Flageole H, Bensoussan AL, Nguyen VH, Laberge JM. Gastrointestinal duplications. *J Pediatr Surg* 2003; **38**: 740-744 [PMID: 12720184 DOI: 10.1016/jpsu.2003.50197]
7 **Correia-Pinto J**, Romero R, Carvalho JL, Silva G, Guimarães H, Estevão-Costa J. Neonatal perforation of a Y-shaped sigmoid duplication. *J Pediatr Surg* 2001; **36**: 1422-1424 [PMID: 11528620 DOI: 10.1053/jpsu.2001.26390]
8 **Salvador II**, Modelli ME, Pereira CR. [Tubular duplication of the colon: a case report and review of the literature]. *J Pediatr (Rio J)* 1996; **72**: 254-257 [PMID: 14688938 DOI: 10.2223/jped.628]
9 **Fotiadis C**, Genetzakis M, Papandreou I, Misiakos EP, Agapitos E, Zografos GC. Colonic duplication in adults: report of two cases presenting with rectal bleeding. *World J Gastroenterol* 2005; **11**: 5072-5074 [PMID: 16124070 DOI: 10.3748/wjg.v11.i32.5072]
10 **Nolan HR**, Wengler C, Hartin CW, Glenn JB. Laparoscopic excision of an ascending colon duplication cyst in an adolescent. *J Pediatr Surg Case Rep* 2016; **4**: 32-34 [DOI: 10.1016/j.epsc.2015.11.007]
11 **Kang M**, An J, Chung DH, Cho HY. Adenocarcinoma arising in a colonic duplication cyst: a case report and review of the literature. *Korean J Pathol* 2014; **48**: 62-65 [PMID: 24627698 DOI: 10.4132/KoreanJPathol.2014.48.1.62]
12 **Heiberg ML**, Marshall KG, Himal HS. Carcinoma arising in a duplicated colon. Case report and review of literature. *Br J Surg* 1973; **60**: 981-982 [PMID: 4764750 DOI: 10.1002/bjs.1800601218]
13 **Jung KH**, Jang SM, Joo YW, Oh YH, Park YW, Paik HG, Choi JH. Adenocarcinoma arising in a duplication of the cecum. *Korean J Intern Med* 2012; **27**: 103-106 [PMID: 22403508 DOI: 10.3904/kjim.2012.27.1.103]
14 **Lee J**, Jeon YH, Lee S. Papillary adenocarcinoma arising in a duplication of the cecum. *Abdom Imaging* 2008; **33**: 601-603 [PMID: 17912582 DOI: 10.1007/s00261-007-9330-1]
15 **Shanmugalingam A**, Duxbury H, Choi JDW, Kwik C, P'Ng CH, Kim L, Pathma-Nathan N. An unusual case of colonic duplication cyst in an adult with dysplasia. *J Surg Case Rep* 2023; **2023**: rjad039 [PMID: 36824693 DOI: 10.1093/jscr/rjad039]
16 **Radhakrishnan L**, George J, Abraham LK. Right-Sided Colonic Duplication Cyst with a Malignant Twist in a Young Adult - a Case Report. *J Gastrointest Cancer* 2022; **53**: 805-808 [PMID: 34279795 DOI: 10.1007/s12029-021-00671-5]
17 **Hsu H**, Gueng MK, Tseng YH, Wu CC, Liu PH, Chen CC. Adenocarcinoma arising from colonic duplication cyst with metastasis to omentum: A case report. *J Clin Ultrasound* 2011; **39**: 41-43 [PMID: 20812340 DOI: 10.1002/jcu.20739]
18 **Carachi R**, Azmy A. Foregut duplications. *Pediatr Surg Int* 2002; **18**: 371-374 [PMID: 12415358 DOI: 10.1007/s00383-002-0835-y]
19 **Yan J**, Yan J, Ding C, Guo J, Peng Y, Chen Y. Clinical features of colorectal duplication in children: A study of 25 cases. *J Pediatr Surg* 2022; **57**: 97-101 [PMID: 34706815 DOI: 10.1016/j.jpedsurg.2021.09.042]

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