

CASE REPORT

Colonic Duplication Cyst Mimicking as a Cystic Pancreatic Tumour: Case Report and Review

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ABSTRACT

Cystic mass presenting in the right hypochondrium, could be a diagnostic challenge. These lesions could arise from pancreas, common bile duct, liver, stomach and colon. Establishing a preoperative diagnosis with certainty, despite the modern radiological imaging and endoscopic facility may be difficult, particularly when it is due to an uncommon lesion like colon duplication cyst. We present here a case of a young lady who presented with one year history of abdominal pain. computerized tomography and magnetic resonance imaging scan revealed a large cystic mass, medial to the "C" loop of duodenum, mimicking a possible cystic lesion of the pancreas. Resection of the lesion and histopathology revealed a duplication cyst of the transverse colon. We discuss the various differential diagnosis of cystic lesion in the right hypochondrium and highlight the salient features of colonic duplication cyst.

INTRODUCTION

Cystic lesions in the right hypochondrium often present as a clinical problem. Several probabilities arise, including the relatively common lesions related to the pancreas [1, 2], cystic lesions of the liver [3] and the uncommon choledochal cyst arising from the common bile duct [4]. However, duplication cysts from the stomach [5, 6] or transverse colon [6] presenting as a cystic right hypochondriac mass is extremely uncommon and is a diagnostic enigma. We present here a case of duplication cyst of the transverse colon in an adult, mimicking a pancreatic lesion and discuss the differential diagnosis. In addition, we highlight the pathology, diagnostic challenge and management issues related to duplication cyst arising from the transverse colon.

Case report

A thirty-eight-year-old female patient was referred to our unit with one year history of chronic upper abdominal pain. Her appetite was good, she had no weight loss and her bowel habit was normal. She was not febrile nor was she jaundiced. On examination, she was not anemic and her vital signs were normal. Abdominal examination revealed a non, tender, non-mobile, large mass in the right hypochondrium. There was no organomegaly.

Her complete blood picture (CBC), liver function tests (LFT), serum amylase and urea electrolytes results were unremarkable. CT and MRI scan of the abdomen revealed a 9x7 cm cystic mass, arising medial to the duodenal loop and adherent to the pancreas, greater curvature of stomach and hepatic flexure. It had few fine septations (**Figure 1-3**). The CBD and the pancreatic duct were normal and the cyst was not communicating to them. Differential diagnosis of pseudocyst of pancreas, mucinous cystadenoma and gastric or colonic duplication cyst were entertained. She was explored with right subcostal incision and findings consistent with CT findings were observed. The cyst was decompressed to facilitate dissection and 200 ml of amber coloured fluid was aspirated (**Figure 4**). The cyst was gradually separated from the duodenum, head of the pancreas and greater curvature of the stomach. However as it seemed to arise from the colon and being close to the hepatic flexure, a right hemicolectomy was carried out (**Figure 5&6**). The histopathology revealed cystic lesion arising from the colon with features consistent with benign cyst with spindle cell proliferation with inflammation and sclerosing fibrosis and areas of smooth muscle. Patient had an uneventful recovery and was discharged on the 7th post-operative day.

DISCUSSION

Cystic mass in the right hypochondrium, could arise from different organs in that region, of which pancreas is the most likely. Several cystic lesions of the pancreas could be considered in the differential diagnosis, based on the patients age, clinical presentation and radiological findings [8-11]. The most predominant differential diagnosis include IPMN, mucinous cystadenoma, serous cystadenoma, simple cysts and pseudocysts related to acute or chronic pancreatitis [1-2, 8-11]. IPMN is characterized by cystic

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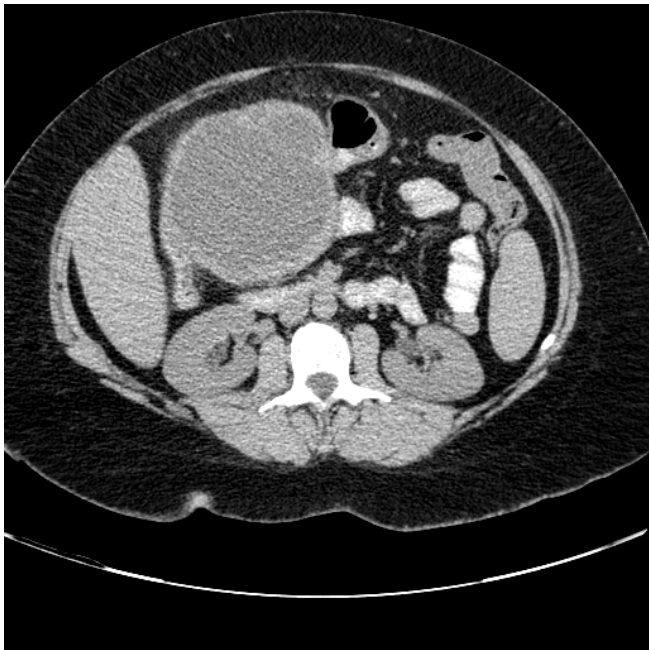


Figure 1. CT scan revealing a cystic lesion medial to the duodenal loop. Fine septations seen on the superomedial aspect of the cyst

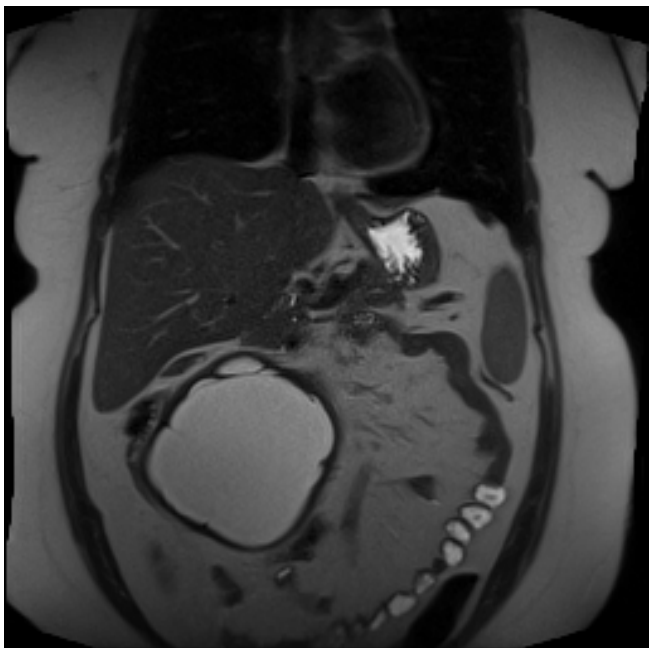


Figure 2. MRI- T2 weighted - coronal view revealing a large cystic lesion in the right hypochondrium

dilation of pancreatic ducts, in which an intraductal proliferation of neoplastic mucin producing cells is usually arranged in papillary pattern [8]. They could arise from main duct, branch duct or both ducts (combined form) and have a risk of turning malignant in 46 to 92% of the cases, based on the duct involved [8]. The papillae may range from microscopic to nodular masses [8]. The mucin production by the neoplastic cells is usually associated with intraluminal mucin secretion, which leads to cystic dilatation of the ducts. When in excess, mucin may drain from the ampulla of Vater, a finding that is virtually diagnostic of IPMN [8, 11]. They generally present in the seventh to eighth decade of life with non specific abdominal symptoms. Mucinous cystic neoplasms share various

features with IPMN [9]. However, in contrast to IPMN, which are cystic dilatations of the pre-existing ducts, MCN's presumably form de novo cystic tumours [9]. They are characterised by an underlying ovarian type of stroma and could turn malignant [9]. Mucinous cystic neoplasms have distinctive clinicopathologic characteristics, including being exclusively seen in perimenopausal females (mean age 48 years; male to female ratio <1/20). The neoplasms is most often located in the tail of the pancreas [9]. Macroscopically, MCN's are composed of large multilocular cysts, which have thick fibrotic wall. Serous cystadenoma on the other hand are benign neoplasm composed of uniform cuboidal, glycogen rich epithelial cells, typically forming innumerable small cysts containing serous fluid, giving a sponge like gross appearance [10]. They usually present as relatively large masses, mostly in the body or tail of the pancreas and are seen predominately in females (male to female ratio; 1:3). The mean age of patients is 61 years and are invariably benign in nature [9]. Pseudocysts, that are much more common than the cystadenomas, are likely to be preceded by a history of acute or chronic pancreatitis [8]. Radiological imaging and analysis of the aspirated fluid for amylase, carcino embryonic antigen (CEA), mucin and malignant cells, could differentiate between these pancreatic lesions [8, 11]. High amylase levels in the aspirated fluid would suggest pseudocyst. Endoscopic ultrasound (EUS) for presence of mural nodules and cyst fluid analysis for cytology, extracellular mucin and CEA levels may help in establishing the diagnosis as these would be elevated in mucinous cystadenoma and IPMN [8, 11]. Radiological/endoscopic investigations that confirm the presence of ductal communication of the cysts, in addition to the presence of papillary process and intraductal mucin, is consistent with the diagnosis of IPMN

Cystic lesions of the liver could also present as right hypochondriac mass. Cystic hepatic lesions could be developmental, inflammatory, neoplastic and trauma related lesions. The most commonly encountered lesions are hepatic cysts, which are developmental in nature and are seen in 2.5% of the general population [3]. They are most often asymptomatic and are incidental findings and need no treatment unless complicated by infection or are symptomatic [3]. US will reveal a well marginated, anechoic structure with enhancement of the posterior wall and increased through transmission. On CT and MRI, simple cysts have attenuation of (0-15 HU) and reveal T1 hypointensity and T2 hyperintensity [3]. Other developmental lesion, like biliary hamartoma, are uncommon [3]. Caroli disease on CT scan or MRI shows multiple cystic lesions in the liver with enhancing "central dot sign", which is the portal radicle. Moreover, communication with the biliary tree may be revealed by MRI or ERCP. Inflammatory cystic lesions are generally preceded by a history of fever, are acute in nature and are associated with suggestive clinical signs and leukocytosis. Pyogenic abscess show complex cyst with enhancing rim and amoebic abscess show complex cyst with double target appearance due to a ring of odema around the well

enhancing inflamed wall [3]. To differentiate between pyogenic and amoebic abscess may be difficult and is made based on the clinical and serological findings. Hydatid cyst may show complex cyst with peripheral daughter cysts [3]. Neoplastic cysts are generally biliary cystadenoma and cystadenocarcinoma and are characterised by large complex cystic lesions with enhancing septations. Biopsy would confirm the diagnosis. Cystic HCC are also complex lesions with hypervascular component with washout in portal venous phase [3]. The diagnosis could be established by elevated tumour markers like alpha feto protein and biopsy. Choledochal cysts are other uncommon cystic lesions that may appear as right hypochondriac cystic mass⁴. They are more common in women and occur in adults in about 20% of the cases, between 20 and 30 years [4]. They may present with pain and when complicated, with fever and jaundice. US, MRCP and if needed ERCP would confirm the diagnosis [4].

Enteric duplication cysts on the other hand are uncommon congenital malformations [11-19]. Less than 50 colonic duplication cysts have been reported so far of which about eighteen are from transverse colon [12]. They are usually diagnosed in infancy and childhood (80% before 2 years), but may be detected at any period of life [16]. They could occur anywhere along the length of the alimentary tract, on the mesenteric side. The most frequent localization is in the ileum 30% and the ileocaecal valve 30%, followed by jejunum 8%, colon 6% to 7% and the rectum 5% [12, 13]. They are characterized by attachment to at least one part of the tract. The malformation is formed before differentiation of the lining epithelium and are therefore named, based on which part of the bowel they arise from, rather than their mucosal lining [12]. Four varieties have been described depending on the shape, including (1) tubular structure that branches from the intestine and extends for some distance between the mesenteric leaves; (2) a double barrel structure communicating with the intestinal lumen at one or both ends; (3) a cystic structure lying within the peritoneal cavity attached by a mesenteric stalk; and (4) a spherical lesion contiguous with some part of the bowel, particularly along the ileum [12]. Cystic types are the most frequent and are seen in 90% to 95% of these cases, while the tubular type in the remaining 5% to 10% [12, 21]. While duplication cysts typically do not communicate with the adjacent bowel lumen, tubular lesions which usually arise near the colon, may communicate [12, 14, 15].

Duplication cyst arising from transverse colon is extremely rare and when they occur on the right side could be a part of the plethora of differential diagnosis. Their walls may contain all of the normal bowel layers, including the mucosa, submucosa and muscularis¹⁴. The cystic type of the transverse colon duplication cysts may have fluid inside it and could be adherent to the mesocolon and the pancreas, mimicking a pancreatic tumour as in our patient (Figure 3) [12].

Most colonic duplication cysts are asymptomatic and remain undiagnosed for years [12-19]. If symptomatic,

they may present with abdominal mass, chronic abdominal pain, constipation, and less frequently with obstruction (volvulus, intussusception), bleeding, infection or perforation [12-19]. The symptoms like abdominal pain may depend on the type of duplication cyst and the associated abnormality. Carcinoma arising from it is another concern [18]. It is difficult to diagnose duplication cyst clinically [12, 14, 15]. Radiological studies play an important role in detection and diagnosis of the duplication cysts [12, 20, 22-24]. Plain radiography may be normal or may show a soft tissue mass with/ without displacement of adjacent bowel or evidence of intestinal obstruction [14]. Ultrasound is particularly well suited for the identification and characterisation of duplication cysts [23, 24]. US of the duplication cyst are characterized by "gut signature" which refers to the hyperechoic inner layer produced by the mucosa surrounded by hypoechoic outer layer caused by smooth muscle [23]. Peristalsis of the cyst wall has also been noted in US [24]. Examinations of the gastrointestinal tract can be useful in order to demonstrate displaced loops of bowel, surrounding the presumed cyst and may delineate the communication with the transverse colon. Contrast barium enema may show communication between the duplication cyst and the normal bowel, with opacification of the two colons [24]. CT and MRI are useful in delineating the relationship between

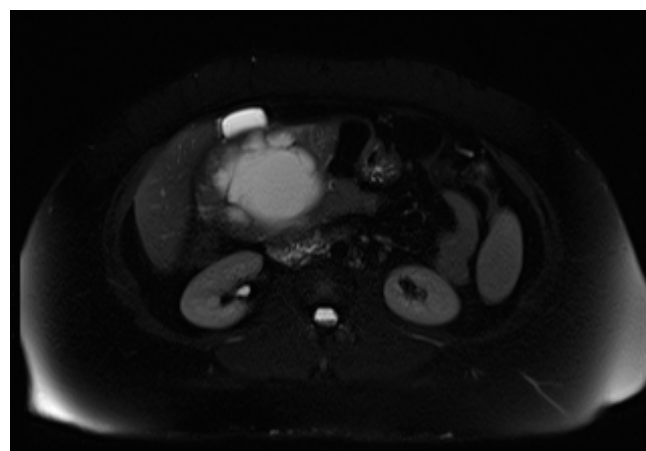


Figure 3. MRI transverse view-T2 weighted revealing a cystic lesion in relation to the transverse colon

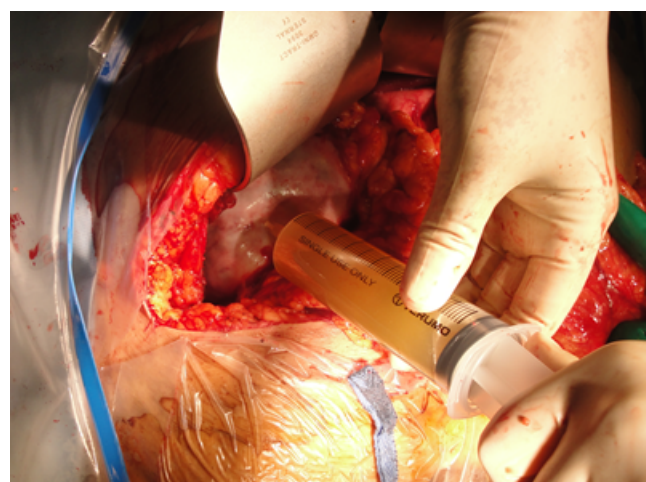


Figure 4. Aspiration of the amber coloured fluid from the cyst

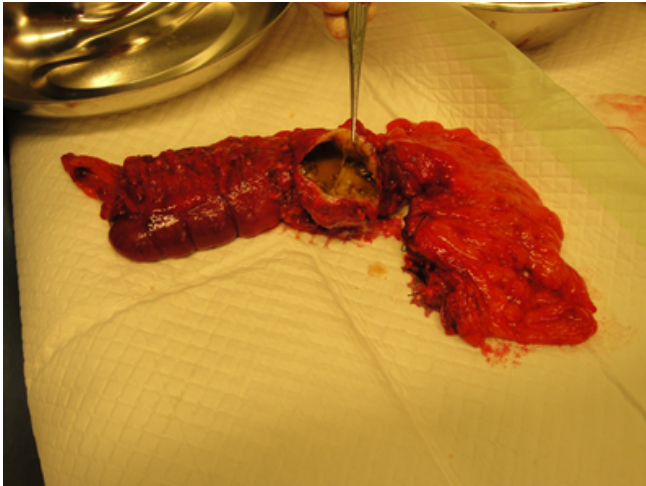


Figure 5. The opened up duplication cyst in relation to transverse colon

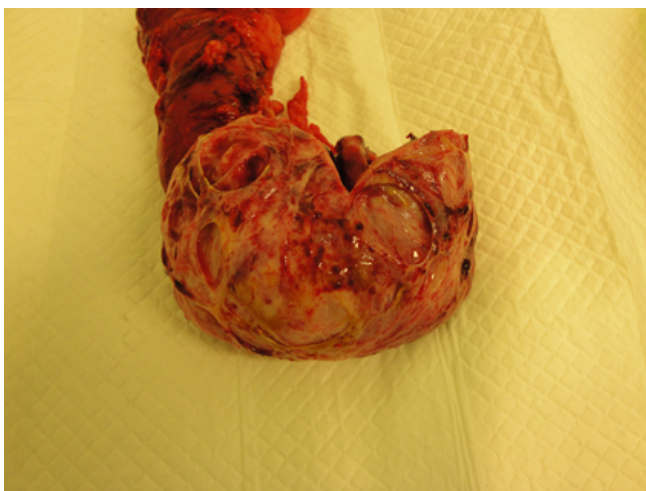


Figure 6. Opened up cyst revealing few septations and inflammation of cyst wall lining

cystic lesion and peripheral structures [12, 14, 16]. In the tubular type, CT scan, contrast enema and colonoscopy could be the best diagnostic tool. The presence of air and stool within the cyst could facilitate the diagnosis [12]. Occasionally, communication between pancreatic duct and duplication cyst may be seen on endoscopic retrograde cholangiopancreatography [25]. Technetium scan may demonstrate ectopic gastric mucosa in the duplication cyst [26].

The recommended treatment of duplication cyst is resection [12-19]. This is not only to relieve the symptomatic patients of their symptoms but also to avoid future complications like obstruction, perforation and bleeding [12-19]. Resection of the duplication cyst is always accompanied by resection of part of the adjacent bowel because of the possibility of malignant changes and the risk of gastrointestinal ulceration and haemorrhage due to ectopic gastric mucosa [12]. When the cyst is long standing, there is a possibility of adhesions to surrounding structures. While one may be able to separate these reasonably easily on most of the occasions, at times because of the intimate attachments of the common wall or because isolated resection may compromise blood flow to the adjacent intestinal segment, resection of adjoining

adherent structure including bowel may be warranted [12]. A frozen section may be carried out to establish the diagnosis or to rule out an occasional concern of lingering malignancy [12].

CONCLUSIONS

Duplication cyst of the colon should be considered in the differential diagnosis of right hypochondriac cystic masses, particularly when the diagnosis is not obvious, despite several investigations. As most of them present in children, its rarity in adult makes a preoperative diagnosis difficult to be established with certainty. The duplication cyst once diagnosed should be resected both in symptomatic and in asymptomatic patients because of its propensity to present with complications, including malignancy. Most of the cystic lesions in the right hypochondriac region including those arising from pancreas, liver and CBD can be diagnosed by modern radiological modalities. Endoscopy and biochemical /microscopic assessment of the fluid aspirate would further facilitate this. However duplication cyst could still spring a surprise because of its rarity and nonspecific presentation and imaging findings.

Conflict of Interest

The authors declare that they have no conflicts of interest concerning this article.

References

1. Nelsen EM, Buehler D, Soni AV, Gopal DV. Endoscopic ultrasound in the evaluation of pancreatic neoplasms-solid and cystic: A review. *World J Gastrointest Endosc* 2015; 7:318-27. [PMID: 25901210]
2. Hocke M, Cui XW, Domagk D, Ignee A, Dietrich CF. Pancreatic cystic lesions: The value of contrast-enhanced endoscopic ultrasound to influence the clinical pathway. *Endosc Ultrasound* 2014; 3:123-30. [PMID: 24955342]
3. Borhani AA, Wiant A, Heller MT. Cystic hepatic lesions: a review and an algorithmic approach. *AJR Am J Roentgenol* 2014; 203:1192-204. [PMID: 25623323]
4. Soares KC, Arnaoutakis DJ, Kamel I, Rastegar N, Anders R, Maithel S, Pawlik TM. Choledochal cysts: presentation, clinical differentiation, and management. *J Am Coll Surg* 2014; 219:1167-80. [PMID: 25442379]
5. D'Journo XB, Moutardier V, Turrini O, Guiramand J, Lelong B, Pesenti C, Monges G, Giovannini M, Delperro JR. Gastric duplication in an adult mimicking mucinous cystadenoma of the pancreas. *J Clin Pathol* 2004; 57:1215-8. [PMID: 15509688]
6. Glaser C, Kuzinkovas V, Maurer C, et al. A large duplication cyst of the stomach in an adult presenting as pancreatic pseudocyst. *Dig Surg* 1998; 15:703-6. [PMID: 9845641]
7. Carneiro FP, de Nazareth Sobreira M, de Azevedo AE, Alves AP, Campos KM. Colonic duplication in an adult mimicking a tumor of pancreas. *World J Gastroenterol* 2008; 14:966-8. [PMID: 18240361]
8. Machado NO, Qadhi H, Wahibi K. Intraductal papillary mucinous neoplasm of pancreas. Understanding of an evolving disease. *Review. N Am J Med Sci* 2015; 7:23-32. [PMID: 20362793]
9. Fukushima N, Zamboni G. Mucinous cystic neoplasms of the pancreas: update on the surgical pathology and molecular genetics. *Semin Diagn Pathol* 2014; 31:467-74. [PMID: 25441310]

10. Reid MD, Choi H, Balci S, Akkas G, Adsay V. Serous cystic neoplasms of the pancreas: clinicopathologic and molecular characteristics. *Semin Diagn Pathol* 2014; 31:475-83. [PMID: 25441309]
11. Nelsen EM, Buehler D, Soni AV, Gopal DV. Endoscopic ultrasound in the evaluation of pancreatic neoplasms-solid and cystic: A review. *World J Gastrointest Endosc* 2015; 7: 318-327. [PMID: 25901210]
12. 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; 19:586-9. [PMID: 23382641]
13. 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]
14. Stefanidis K, Lappas I, Kolofousi C, Kalogeropoulos I. A rare presentation of colonic duplication cyst: report of a case and review of literature. *JBR-BTR* 2012; 95:71-3. [PMID: 22764658]
15. Ademuyiwa AO, Bode CO, Adesanya OA, Elebute OA. Duplication cyst of ascending colon presenting as an ileal volvulus in a child: a case report and review of literature. *Afr J Paediatr Surg* 2012; 9:237-9. [PMID: 23250247]
16. Nichols KC, Pollema T, Moncure M. Laparoscopically excised completely isolated enteric duplication cyst in adult female: a case report. *Surg Laparosc Endosc Percutan Tech* 2011; 21:e173-5. [PMID: 21857453]
17. Mourra N, Chafai N, Bessoud B, Reveri V, Werbrouck A, Tiret E. Colorectal duplication in adults: report of seven cases and review of the literature. *J Clin Pathol* 2010; 63:1080-3. [PMID: 20924093]
18. 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; 39:41-3. [PMID: 20812340]
19. Cavallaro G, Arena R, D'Ermo G, Basile U, Polistena A, Scorsi A, Mingazzini PL, De Toma G. Cystic duplication of transverse colon: an unusual case of abdominal pain and bowel obstruction. *G Chir* 2010; 31:236-8. [PMID: 20615367]
20. Inoue Y, Nakamura H. Adenocarcinoma arising in colonic duplication cysts with calcification: CT findings of two cases. *Abdom Imaging* 1998; 23:135-7. [PMID: 9516499]
21. Dutheil-Doco A, Ducou Le Pointe H, Larroquet M, Ben Lagha N, Montagne J. A case of perforated cystic duplication of the transverse colon. *Pediatr Radiol* 1998; 28:20-2. [PMID: 9426268]
22. Pintér AB, Schubert W, Szemlédy F, Göbel P, Schäfer J, Kustos G. Alimentary tract duplications in infants and children. *Eur J Pediatr Surg* 1992; 2:8-12. [PMID: 1571331]
23. Barr LL, Hayden CK Jr, Stansberry SD, Swischuk LE. Enteric duplication cysts in children: are their ultrasonographic wall characteristics diagnostic? *Pediatr Radiol* 1990; 20: 326-328. [PMID: 2190153]
24. Spottswood SE. Peristalsis in duplication cyst: a new diagnostic sonographic finding. *Pediatr Radiol* 1994; 24:344-345. [PMID: 7824371]
25. Rathi V, Singh S, Bhargava SK, Kaur N, Seth N. Diagnosis of tubular colonic duplication by barium follow-through study. *Australas Radiol* 2005; 49:157-159. [PMID: 15845056]
26. Luckmann KF, Welch RW, Schwesinger W, Oswalt C, Bannayan G. Symptomatic duodenal duplication cyst in an adult demonstrated by endoscopic retrograde cholangiopancreatography. Case report and literature review. *Am J Gastroenterol* 1979; 72:153-159. [PMID: 474555]
27. Emamian SA, Shalaby-Rana E, Majd M. The spectrum of heterotopic gastric mucosa in children detected by Tc-99m pertechnetate scintigraphy. *Clin Nucl Med* 2001; 26: 529-535. [PMID: 11353301]