Journal of Pediatric Surgery Case Reports 64 (2021) 101723

Contents lists available at ScienceDirect

Journal of Pediatric Surgery Case Reports

journal homepage: http://www.elsevier.com/locate/epsc

Gastric duplication presenting as partial gastric outlet obstruction

Giulia Fusi [a,b], Francesco Molinaro [a,b], Francesco Ferrara [b], Alessandra Taddei [b], Franco Roviello [c], Luigi Marano [c], Fabiola Rossi [d,e], Maurizio Costantini [e], Alessandro Cappelli [f], Mario Messina [a,b], Rossella Angotti [b,*]

[a] *Division of Pediatric Surgery, Department of Medical, Surgical and Neurological Sciences, University of Siena, Siena, Italy*
[b] *Division of Pediatric Surgery, Department of Women and Children, Le Scotte Hospital, Siena, Italy*
[c] *General Surgery and Surgical Oncology Department, University of Siena, Italy*
[d] *Division of Human Pathology and Oncology, Department of Medical Biotechnology, University of Siena, Siena, Italy*
[e] *Department of Human Pathology and Oncology, University of Siena, Siena, Italy*
[f] *Vascular and Endovascular Surgery Unit, Department of Medicine, Surgery, and Neuroscience, University of Siena, Siena, Italy*

ARTICLE INFO

Keywords:
Bowel duplication
Abdominal pain
Child

ABSTRACT

We present a very rare case of gastric duplication treated with complete excision with two surgical procedures. We decided to report our case to share our experience that confirm the difficult of preoperative/intraoperative diagnosis of gastric duplication in a pediatric patient. Finally we reviewed the literature to date.

1. Introduction

Gastric duplication cyst (GDC) is an extremely rare malformation (2–9%) [1]. It is mostly diagnosed in infancy and only a few cases are reported in Literature. The preoperative diagnosis is a challenge for pediatric surgeon, because it often onset as non-specific acute abdomen. We report a case of complicated GDC in a 3 years-old female, who came to our attention for abdominal pain associated with non bilious vomiting since 2 days. Due to the risk associated with radiation based on the age of child and the suspicion of intussusception by ultrasound, we performed a first diagnostic laparoscopy. After two weeks an elective surgery was performed with complete excision of duplication. We also present a review of the literature of similar cases.

2. Case report

A 3 years old female was transferred to our hospital with a suspicion of bowel intussusception. She presented a diffuse and colicky abdominal pain associated with non bilious vomiting (15 episodes) since 2 days. No others symptoms associated (fever, diarrhoea, decreased urine output, decreased appetite, weakness and fatigue). No Bright red rectal bleeding mixed with mucus. The child was well. The physical examination showed a soft abdomen and a palpable mass in the epigastrium. Ultrasonography showed absence of imaging of intussusception. Based on clinical picture of child we decided for an active clinical observation. After few hours, child become pale, lethargic, tachycardic. Abdomen became tenderness with evidence of peritonism. Due to the clinical evolution, we decided to perform a laparoscopy. The procedure became diagnostic in absence of emergency condition and definitive diagnostic. It showed only a diffuse retroperitoneal hematoma. CT scan was performed on POD 3 but it confirmed a diffuse retroperitoneal hematoma with signs of active bleeding (Fig. 1) and no clear anomalies were found. We completed the diagnostic workup with an endoscopic ultrasonography that showed a regular gastro-duodenal region and an MRI that confirmed a duodenal cyst as duplication. We have planned an elective laparotomy after 2 weeks from the first laparoscopy. Gastroduodenal region was exposed and the cyst (4 × 3 cm) was found arising from pylorus. An intraoperative gastroscopy was done to confirm the absence of communication with stomach and to preserve the common Wall during Excision of the gastric Duplication. Gastric duplication was removed “en bloc” (Fig. 2). It contained mucinous fluid and presented ulcerative *trans*-mural area in the posterior wall. Histopathology confirmed a gastric duplication with heterotopic pancreatic and gastric tissue, perforation and peritonitis (Fig. 3). Postoperative course was uncomplicated. The patient was discharged home 11 days later. A 6 months follow up showed no complication, regular growth and nutrition.

* Corresponding author. Viale Bracci 16, Siena, 53100, Italy.
E-mail address: rossellaangotti@me.com (R. Angotti).

https://doi.org/10.1016/j.epsc.2020.101723
Received 24 October 2020; Received in revised form 15 November 2020; Accepted 17 November 2020
Available online 6 December 2020
2213-5766/© 2020 The Authors. Published by Elsevier Inc. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

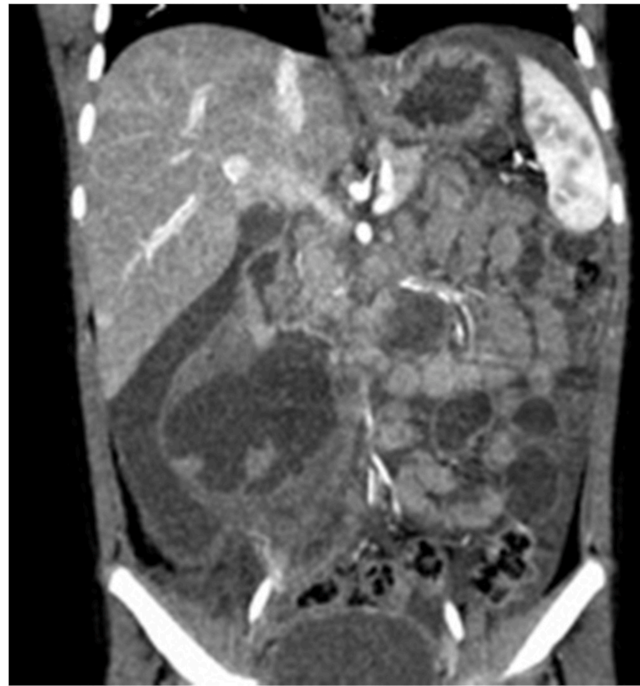

Fig. 1. CT with IV contrast: massive retroperitoneal hematoma with signs of both previous and active bleeding probably due to a leakage from the right gastric artery.

3. Discussion

Duplications of the alimentary tract are rare congenital malformations that may affect each segment of the alimentary tract [1]. Gastric/duodenal and rectal duplications are the rarest [2]. Gastric is involved for only 2–9%, with an incidence rate of 17:1000000 individuals and a major prevalence in female [3]. Gastric duplications are classically divided into tubular and cystic types. Tubular are usually interlinked with the stomach. Cystic-type are the most common and involve the greater curvature of the stomach and non-communicating. Our case conforms to the Literature. GDCs may be asymptomatic but usually become symptomatic before 2 years of age [4]. When GDCs are symptomatic, clinical presentation is totally a-specific and sometimes leads to misdiagnosis. In fact, they may cause various abdominal

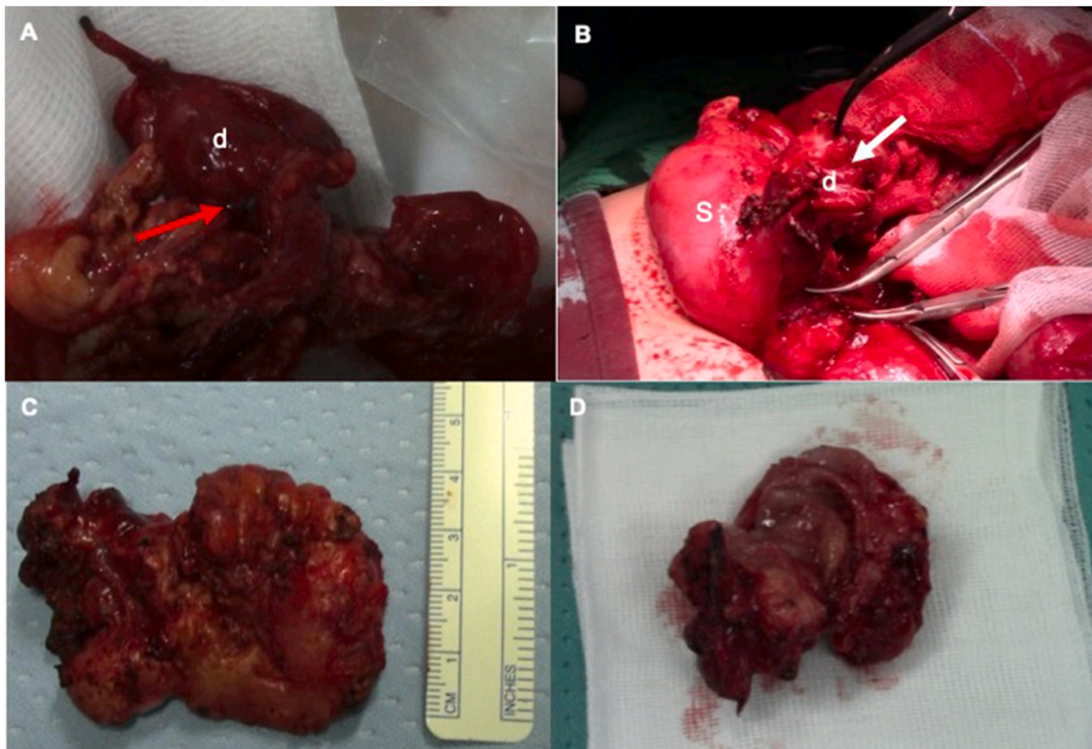

Fig. 2. Intraoperative images. A-B isolated cyst: S (stomach), d (duplication; C-D removal cyst en bloc.

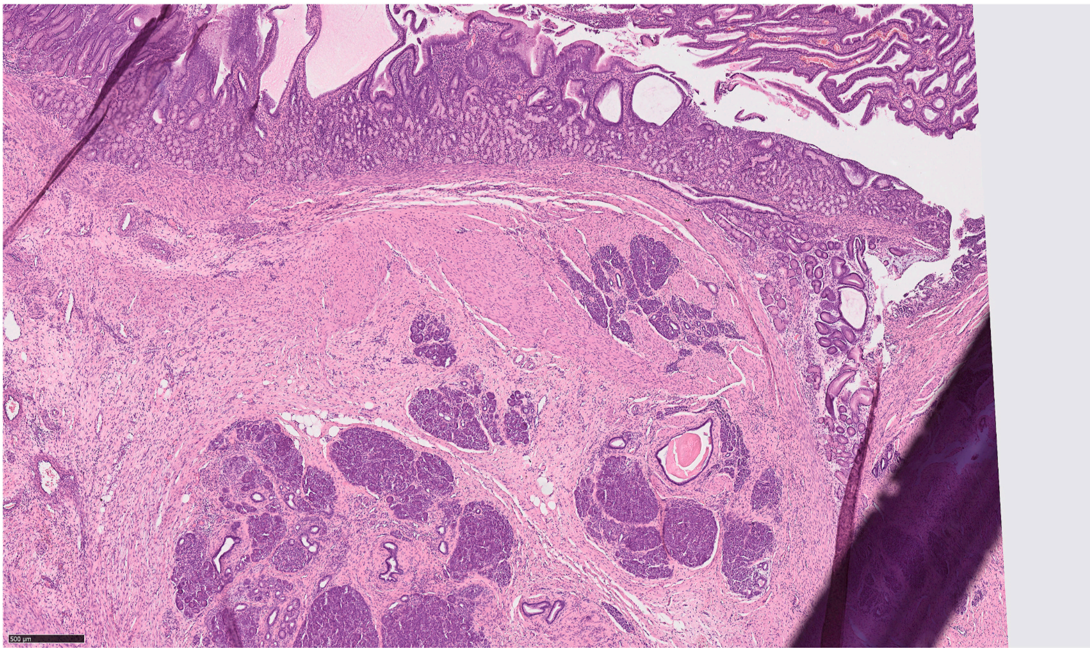

Fig. 3. Histopathology (2X): gastric mucosa (gastric antrum and pylorus) and glands refer to pancreas.

symptoms, from palpable abdominal mass, pain, vomiting, bloody stools, feeding difficulties, loss of weight. Sometimes the presentation can be complicated as an intussusception, volvulus of the small bowel, bleeding or perforation due to the ulceration of ectopic mucosa [3]. The absence of specific symptoms and signs and the difficulty to appropriate imaging, as reported by some Authors, often the surgeon to misdiagnosis. Clinical presentation and imaging may be helpful to suspect diagnosis but it is no enough to confirm it [3]. The Literature reports many cases with clinical features and preoperative imaging results different respect to intraoperative diagnosis [5].

There is no known a gold standard preoperative exam. Ultrasounds or radiographic studies are often used as first line exams but they are aspecific. Esophagogastroduodenoscopy is often no diagnostic because of GDCs are not communicating [3], however sometimes it may be helpful to identify ulcers or strictures, and better define the anatomy of duplication cysts in the upper gastrointestinal tract before to surgery or intraoperative. CT scan and MRI can be used in cases where ultrasound is unclear or in complicated cases but often they are no diagnostic. Furthermore, about ct scan, due to the risk associated with radiation and due to the costs, a selective use is recommended. About the MRI, should be considered that it may not be used always in emergency and it often need of sedation. Endoscopic ultra sound is a new diagnostic tool, that combines the advantages of ultrasound and esophagogastroduodenoscopy. It may be useful to distinguish between interlinked and no interlinked gastric duplications [6]. However, also this imaging, is often affected by the presence of collateral findings that can cause artefact (blood, inflammatory tissue), like our case. In the landscape of imaging available for the preoperative diagnosis of GDC, the MRI certainly plays an important role after ultrasound. However, GDCs are often diagnosed during surgery [7]. Our case shows exactly how pre-operative and intraoperative diagnosis of gastric duplication cyst (GDC) can be challenging. CT scan and MRI were invalidated by bleeding. About the management of GDC there is a shared attitude between pediatric surgeons in case of complicated onset but it remains controversial in case of asymptomatic patient. In complicated clinical picture, indeed, is imperative a surgical approach to manage the emergency; sometimes it can be definitive, sometimes only diagnostic or partially definitive. In asymptomatic child, instead, the choice of surgery is no clear. It is certain mandatory a large and thorough discussion with the parents [8]. They should be informed about the possible complications and the potential but rare risk of neoplasia in adult age [6]. Because it is impossible to prevent the future evolution and sometimes risks of surgery are high, a tight and a long term follow up (clinical and radiologist) could be a good option. In case of elective surgery a complete removal of the duplication is the recommendable choice. It is conceivable, however, the possibility of partial gastrectomy or mucosal stripping [8] in case of large gastric duplication. Several Authors suggest the use of minimally invasive techniques for the excision of GDCs [8]. Laparoscopic or robotic approaches [3,8] are reported in Literature with high index of success, but they are available for simple and no complicated cyst. Laparotomy remains the only choice, like in our case, in complicated cases with multiple adhesions and the presence of abundant chronical inflammatory tissue. In this case, indeed, the mininvasive approach could be dangerous for patient. The two stage approach as in our case, is a no common option but it has been reported also by Callahan [10]. As these Authors, indeed, we did not identify the gastric duplication by preoperative imaging and by first surgical procedure and we focused our attention about the trauma or abuse of patients as cause of retroperitoneal hematoma. Our experience confirms that the diagnosis of gastric duplication is a challenge for paediatric surgeon. We reviewed the Literature and we found 47 reported patients as it is shown in Table 1 [11–35].

4. Conclusions

Based on our experience, we can conclude that GDC should be considered a possible diagnosis in patients with no specific abdominal pain and non bilious vomiting and not clear preoperative imaging, especially if under 3 years. The presence of retroperitoneal bleeding, furthermore, must be considered a high index of suspicion of a complicated gastro-duodenal duplication. Surgery is imperative and resolutive. The mininvasive approach can be considered safe procedure when done by skilled surgeons and in no complicated case.

Patient consent

Consent to publish the case report was not obtained. This report does not contain any personal information that could lead to the

Table 1
Case series of gastric duplication cysts reported in pediatric literature in the last 10 years (2009–2019).

Author (year)	Case	Age/NA (not available)	Symptoms/NA (not available)	Urgent/elective surgery/NA (not available)	Open (O); MIS (VLS/Robot); Endoscopy (E); NA (not available)	Type of surgery
Upadhyaya VD et al., 2009 [11]	1	21 d	Abdominal mass, non bilious vomiting	URGENCY	OPEN	Cystectomy and resected of pyloroduodenal region. and end to end anastomosis
Chattopadhyay et al., 2010 [12]	2	8 m	Asymptomatic	ELECTIVE	OPEN	Cystectomy
Biebl et al., 2010 [13]	3	8 y	Abdominal pain, non bilious vomiting	URGENCY	VLS	Partial gastrectomy
Stock et al., 2010 [14]	4	2 y	Abdominal pain, non bilious vomiting	NA	NA	NA
Laje et al., 2010 [15]	5	17 mo	Antenatal diagnosis	ELECTIVE	VLS	Cystectomy
	6	2 m	Antenatal diagnosis	ELECTIVE	OPEN	Cystectomy
	7	3 m	Antenatal diagnosis	ELECTIVE	OPEN	Cystectomy
Kayastha et al., 2010 [16]	8	15 m	Abdominal pain	URGENCY	OPEN	Cystectomy
Chin et al., 2011 [17]	9	11 d	Abdominal pain, non bilious vomiting	URGENCY	VLS	Cystectomy
Prinsloo et al., 2011 [18]	10	3 m	haematemesis	ELECTIVE	VLS→ OPEN	Cystectomy
	11	9 m	Haematemesis/melena	URGENCY	OPEN	Cystectomy
Hartog et al., 2011 [19]	12	1 y	NA	NA	NA	Cystectomy
Lima et al., 2012 [20]	13	1 y	Asymptomatic	ELECTIVE	VLS→ OPEN	Cystectomy
	14	4 y	Abdominal pain	URGENCY	VLS→ OPEN	Cystectomy
	15	1 y	Asymptomatic	ELECTIVE	VLS	Cystectomy
Malays et al., 2012 [21]	16	10 m	Non-bilious vomiting and an abdominal mass	ELECTIVE	OPEN	Cystectomy
Shukla et al., 2012 [22]	17	7 d	Eventration of diaphragm	URGENCY	OPEN	Cystectomy
Surridge et al., 2014 [23]	18	13 m	Rectal bleeding	URGENCY	OPEN	Cystectomy
Okur MH et al., 2014 [24]	19	<2.5 y	NA	NA	NA	NA
Zouari M et al., 2014 [25]	20	<1 y	Acute abdomen	NA	NA	Cystectomy and end-to-end anastomosis
Jehangir et al., 2015 [26]	21	1 <1y	Emesis	URGENCY	OPEN	Cystectomy
	22	1 1y-5y	Emesis	URGENCY	OPEN	Partial gastrectomy
	23	1 1y-5y	Abdominal pain	URGENCY	OPEN	Mucosectomy
Takazawa et al., 2015 [27]	24	2 m	Abdominal pain, non bilious vomiting	URGENCY	VLS	Partial gastrectomy
Jain et al., 2015 [28]	25	5 y	recurrent upper abdominal pain	URGENCY	NA	Cystectomy
Tanaka H et al., 2016 [29]	26	4 y	hypergastrinemia	ELECTIVE	OPEN	Cystectomy
Ren et al., 2017 [30]	27	1 h	Saliva bucking	URGENCY	VLS	Cystectomy
	28	1 d	Emesis	URGENCY	VLS	Cystectomy
	29	26 d	Emesis	URGENCY	VLS	Cystectomy
	30	28 d	Asymptomatic	ELECTIVE	VLS	Cystectomy
	31	24 d	Emesis	URGENCY	VLS	Cystectomy
Balakrishnan et al., 2017 [8]	32	5 Y	Emesis, abdominal pain	URGENCY	VLS	Partial gastrectomy
	33	2 y	Asymptomatic	ELECTIVE	VLS	Cystectomy
	34	10 y	abdominal pain	URGENCY	VLS	Cystectomy
	35	9 y	Emesis, abdominal pain	URGENCY	VLS	Cystectomy
	36	12 y	abdominal pain	URGENCY	VLS	Cystectomy
Koduri B et al., 2017 [31]	37	3 y	vomiting. obstruction and sepsis	URGENCY	OPEN	Cystectomy
Zhang et al., 2017 [3]	38	8y	Abdominal pain, non bilious vomiting	URGENCY	VLS	Cystectomy
	39	20 m	Asymptomatic	ELECTIVE	VLS→ OPEN	Cystectomy
	40	11 y	Recurrent abdominal pain	URGENCY	VLS	Cystectomy
	41	9 m	Melena	URGENCY	VLS	Cystectomy
Tiwari et al., 2017 [32]	42	NA	sickle cell trait, vague abdominal pain, recurrent cough and multiple episodes of haemoptysis.	Na	OPEN	Cystectomy
Rizzo et al., 2018 [9]	43	22 m	asymptomatic	ELECTIVE	Robot	Cystectomy
Arshad M et al., 2018 [33]	44	5 y	gastric outlet obstruction	URGENCY	NA	NA
Lyu TX et al., 2019 [34]	45	7 y	Abdominal pain, non bilious vomiting	URGENCY	VLS	Cystectomy
Güngör T et al., 2019	46	3 m	feeding problems in syndrome	NA	NA	NA
Fang Y ET AL. 2019 [35]	47	5 M	Asymptomatic	ELECTIVE	Endoscopy	Endoscopic submucosal dissection (ESD)
Our case	**48**	**3 y**	**Abdominal pain, non bilious vomiting**	**URGENCY/ ELECTIVE**	**VLS→ OPEN**	Cystectomy

identification of the patient.

Funding

No funding or grant support.

Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

References

[1] Stringer MD, Spitz L, Abel R, et al. Management of alimentary tract duplication in children. Br J Surg 1995;82:74–8.
[2] Molinaro F, Ferrara F, Cerchia E, et al. Rectal duplication cyst in previous anorectal malformation and Down syndrome. Minerva Pediatr 2014;66:654–6.
[3] Zhang L, Chen Q, Gao Z, et al. Diagnosis and treatment of gastric duplication in children: a case report. Experimental and therapeutic medicine 2017;14:3062–6.
[4] Rodriguez CR, Eire PF, Lopez GA, et al. Asymptomatic gastric duplication in a child: report of a new case and review of the literature. Pediatr Surg Int 2005;21:421–2.
[5] Angotti R, Molinaro F, Cobellis G, et al. Persistent nonbilious vomiting in a child: possible duodenal webbing. Clin Endosc 2017;50(2):191–6.
[6] Passos ID, Chatzoulis G, Milias K, et al. Gastric duplication cyst (gdc) associated with ectopic pancreas: case report and review of literature. Int J Surg Case Rep 2017;31:109–13.
[7] Trainavicius K, Gurskas P, Povilavicius J. Duplication cyst of the pylorus: a case report. J Med Case Rep 2013;7:175.
[8] Balakrishanan K, Fonacier F, Sood S, et al. Foregut duplication cysts in children. J Soc Laparoendosc Surg 2017;21(2):e2017.00017.
[9] Rizzo R, Lisi G, Marino N, et al. Robot assisted resection of gastric duplication cysts in a child: a detailed case report. Pediatr Med e Chir 2018;40(2):4.
[10] Callahan K, Lee S, Stewart S, et al. Hemorrhagic pyloroduodenal duplication cyst misdiagnosed as child abuse. J Pediatr 2013;163:1224.
[11] Upadhyaya VD, Srivastava PK, Jaiman R, et al. Duplication cyst of pyloroduodenal canal: a rare cause of neonatal gastric outlet obstruction: a case report. Cases J 2009;12(2):42. 1.
[12] Chattopadhyay A, Mitra SK, Dutta S, et al. Gastric, pancreatic, and ureteric duplication. J Indian Assoc Pediatr Surg 2010;15:25–7.
[13] Biebl M, Hechenleitner P, Renz O, et al. Laparoscopic resection of multiple gastric duplication cysts in an 8 year-old boy. J Pediatr Surg Case Rep 2015;3(3):134–6.
[14] Stock F, Cammarata-Scalisi F, Petrosino P, et al. Gastric duplication. A case report and review of the literature. Acta Gastroenterol Latinoam 2010;40(3):258–63.
[15] Laje P, Flake AW, Adzick NS. Prenatal diagnosis and postnatal resection of intraabdominal enteric duplications. J Pediatr Surg 2010;45(7):1554–8.
[16] Kayastha K, Sheikh A. Gastric duplication cyst presenting as acute abdomen: a case report. APSP J Case Rep 2010;1(1):6.
[17] Chin AC, Radhakrishnan RS, Lloyd J, Reynolds M. Pyloric duplication with communication to the pancreas in a neonate simulating hypertrophic pyloric stenosis. J Pediatr Surg 2011;46:1442–4.
[18] Prinsloo H, Loveland J, Grieve A, et al. Gastric duplication cysts as a rare cause of hematemesis: diagnostic challenges in two children. Pediatr Surg Int 2011;27(10):1127–30.
[19] Hartog H, Dikkers FG, Veldhuizen AG, et al. Cervical cystic swelling in an adolescent: unusual association of a cervical mature teratoma with vertebral anomalies and a history of gastric duplication cyst. J Pediatr Surg 2011;46(6):e15–8.
[20] Lima M, Molinaro F, Ruggeri G, et al. Role of mini-invasive surgery in the treatment of enteric duplications in paediatric age: a survey of 15 years. Pediatr Med e Chir 2012;34(5):217–22.
[21] Malays GKK. Gastric duplication cyst in an infant presenting with non-bilious vomiting. J Med Sci 2012;19(1):76–8.
[22] Shukla RM, Maitra SK, Patra MP, et al. Eventration of diaphragm with gastric duplication cysts: a rare association. Indian J Pediatr 2012;79:1377.
[23] Surridge CA, Goodier MD. Gastric duplication cyst: a rare cause of rectal bleeding in a young child. Afr J Paediatr Surg 2014;11(3):267–8.
[24] Okur MH1, Arslan MS, Arslan S, et al. Gastrointestinal tract duplications in children. Eur Rev Med Pharmacol Sci 2014;18(10):1507–12.
[25] Zouari M, Bouthour H, Abdallah RB, et al. Alimentary tract duplications in children: report of 16 years' experience. Afr J Paediatr Surg 2014;11(4):330–3.
[26] Jehangir S, Ninan PJ, Jacob TJ, et al. Enteric duplication in children: experience from a tertiary center in South India. J Indian Assoc Pediatr Surg 2015;20(4):174–8.
[27] Takazawa S, Uchida H, Kawashima H, et al. Laparoscopic partial gastrectomy of a huge gastric duplication cyst in an infant. Nagoya J Med Sci 2015;77(1–2):291–6.
[28] Jain AS, Patel AM, Jain SR, et al. Accessory pancreatic lobe with gastric duplication cyst: diagnostic challenges of a rare congenital anomaly. BMJ Case Rep 2015;12:2015.
[29] Tanaka H, Masumoto K, Sasaki T, et al. Hypergastrinemia and a duodenal ulcer caused by gastric duplication. Surg Case Rep 2016;2(1):75.
[30] Ren HX, Duan LQ, Wu XX, et al. Laparoscopic resection of gastric duplication cysts in newborns: a report of five cases. BMC Surg 2017;17:37.
[31] Koduri B, McHale K, Yost C, et al. Gastric duplication: a rare cause of Recurrent Vomiting. Case Rep Pediatr 2017;2017:2348274.
[32] Tiwari C, Shah H, Waghmare M, et al. Cysts of gastrointestinal origin in children: varied presentation. Pediatr Gastroenterol Hepatol Nutr 2017;20(2):94–9.
[33] Arshad M, Jeelani SM, Siddiqui A, et al. Duplication cyst of the pylorus: a rare cause of gastric outlet obstruction. BMJ Case Rep 2018;8:2018.
[34] Lyu XT, Pang XL, Wu L, et al. Diagnosis of gastric duplication cysts in a child by endoscopic ultrasonography. Chin Med J (Engl) 2019;132(4):488–90.
[35] Fang Y, Gao T, Yang H, et al. Removal of an infant's gastric duplication cyst through endoscopic submucosal dissection: a case report. Medicine (Baltim) 2019;98(12):e14820.