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Case Report

Surgery Section

Enteric Duplication Cyst in a One-year-old Child: A Case Report

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ABSTRACT

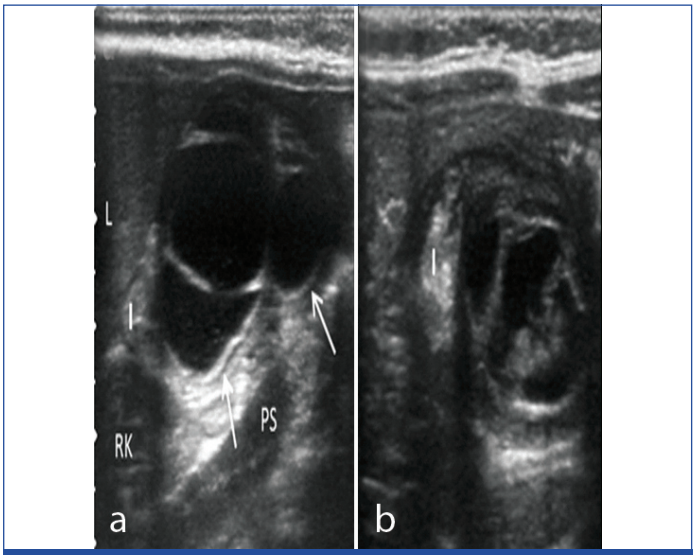
Enteric duplication cysts are rare congenital anomalies of the gastrointestinal tract, predominantly occurring in the ileum, although they can present in various locations, such as the retroperitoneum. These cysts may contain heterotopic tissues and can mimic other cystic lesions of the pancreas and peripancreatic region, posing diagnostic challenges. Hereby, the authors present a case of a one-year-old male child who presented with acute epigastric abdominal pain, vomiting, and fever. Clinical examination revealed a palpable epigastric mass. Ultrasonography (USG) and Contrast-enhanced Computed Tomography (CECT) demonstrated a large, multiloculated cystic lesion in the retroperitoneum, anterior to the pancreas, causing extrinsic gastric compression. Exploratory laparotomy revealed a thick-walled cystic mass adherent to the coeliac artery and splenic vessels, which was completely excised. Histopathological examination confirmed an enteric duplication cyst with a hamartomatous component, comprising multiple epithelial types, pancreatic acini, smooth muscle tissue, and arteriovenous malformations. The present case highlights the importance of considering enteric duplication cysts in the differential diagnosis of retroperitoneal cystic lesions in infants. Complete surgical excision is crucial to prevent potential complications such as infection, perforation, or malignant transformation.

Keywords: Abdominal pain, Congenital abnormality, Cystic lesion, Hamartoma, Laparotomy

CASE REPORT

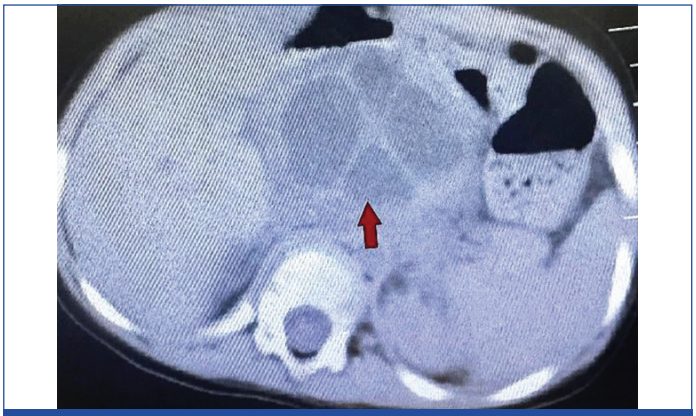
A one-year-old male child presented to the Emergency Department with a two-day history of acute epigastric abdominal pain, vomiting, and fever. The pain was non radiating, aggravated by food consumption, and relieved by lying down. The vomiting was non projectile, non bilious, and comprised gastric contents, occurring two to three times daily. The child also experienced a low-grade fever with chills, which resolved with medication. There was no prior history of similar complaints.

On examination, the abdomen was soft, with a palpable, non tender mass in the epigastric region. There was no guarding or rigidity, and bowel sounds were present. Ultrasonography revealed a well-defined, multiloculated cystic lesion with echogenic content, likely fat, located anterior to the pancreas, with no evidence of internal or peripheral vascularity [Table/Fig-1a,b].



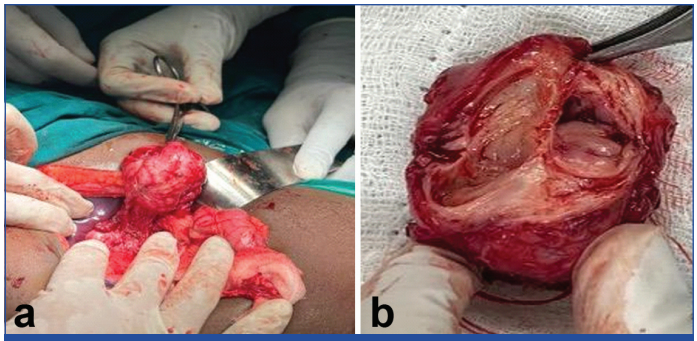
[Table/Fig-1a,b]: Ultrasonography showing multiloculated cystic lesion (white arrows). PS: Pancreas; RK: Right kidney; L: Liver

The CECT of the abdomen and pelvis showed a large, well-defined, multiloculated, thick-walled cystic lesion measuring approximately 45×38×49 mm in the retroperitoneum, posterior and superior to the pancreatic neck and body, extending superiorly into the lesser sac [Table/Fig-2]. It caused extrinsic compression on the body of the stomach and the neck and body of the pancreas, displaying septations and a few calcific foci. There was mild extrinsic compression and anterior displacement of the adjoining common hepatic duct and common bile duct, resulting in mild central dilatation of the intra-hepatic biliary radicles in both the right and left lobes. The splenic vein and portal confluence were displaced anteriorly. The lesion closely abutted the anterior surface of the aorta, inferior vena cava, and the adjoining superior mesenteric artery and superior mesenteric vein. Based on these findings, a provisional diagnosis of pancreatic cyst, mesenteric cyst, or lymphangioma was made. The differential diagnosis included cystic teratoma and mucinous cystadenoma.



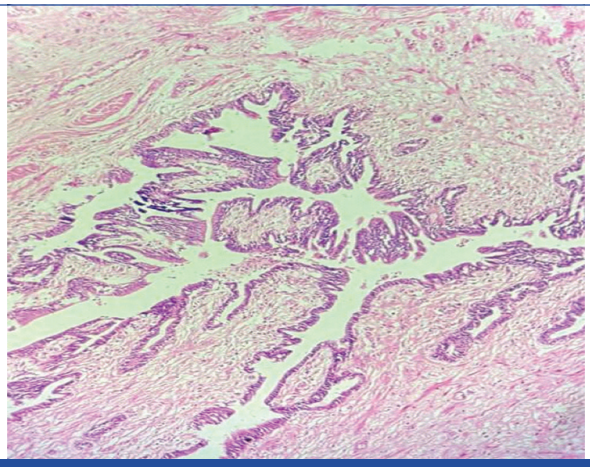
[Table/Fig-2]: The Computed Tomography (CT) image showing multiloculated cystic lesion (red arrow).

The child underwent exploratory laparotomy with a plan for either complete or partial removal of the mass, or biopsy depending on resectability. Intraoperatively, a thick-walled cystic mass was identified in the lesser sac, adherent posteriorly to the coeliac artery and splenic vessels [Table/Fig-3a]. The entire lesion was successfully excised [Table/Fig-3b]. It was a nodular mass measuring 4.2×3.2×2.5 cm. The external surface was greyish-brown and congested, while the cut surface revealed multiloculated cystic spaces with thick grey-white walls. The cyst contained haemorrhagic fluid with a bile tinge.



[Table/Fig-3]: a) Intraoperative image; b) Excised lesion.

Histopathological examination {Haematoxylin and Eosin (H&E)} of the cyst showed a lining of varying epithelial types, including ciliated columnar epithelium, biliary epithelium with goblet cell metaplasia, colonic-type epithelium, and squamous epithelium. The cyst wall contained pancreatic acini, arteriovenous malformations, hypertrophied nerve bundles, abundant smooth muscle tissue, mucin extravasation, and areas of hyalinisation, confirming the diagnosis of an enteric duplication cyst [Table/Fig-4]. Based on clinical, radiological, and primarily histopathological findings, the lesion was diagnosed as an enteric duplication cyst with a hamartomatous component.



[Table/Fig-4]: Histopathology examination (H&E, 400x).

The postoperative period was uneventful. The child was started on a full diet on postoperative day five and discharged on postoperative day eight with no complications. After two months of follow-up, the patient remained asymptomatic.

DISCUSSION

Duplication of the gastrointestinal tract is a rare congenital anomaly, with an incidence of approximately 0.2% in children [1]. These cysts show a higher prevalence in males compared to females [2,3]. While they can present at any age, they are most commonly observed during the first two years of life [1,4]. However, there are cases where patients have presented later, as reported by Bapir R et al., where the patient was 17 years old at presentation, and Hroub O et al., reported a case at 18 years of age [5,6]. The aetiology remains uncertain, but several theories have been proposed, including the split notochord theory (the most widely accepted), partial twinning, persistent embryological diverticula, and aberrant luminal recanalisation [2,7,8]. These anomalies can arise from any part of the alimentary tract, with the ileum being the most frequently affected site (60%), while involvement of the pyloroduodenal, colonic, and rectal regions is rare [9,10].

The clinical presentation of gastrointestinal duplication varies significantly, depending on factors such as location, size, communication with adjacent bowel, and the presence of complications like inflammation [1,11]. Tiwari C et al., conducted a retrospective analysis of 14 patients [12]. The study reported that gastrointestinal cysts present with varied symptoms based on their origin. Common symptoms in infants and neonates include abdominal pain, nausea, vomiting, gastrointestinal bleeding, abdominal distension, the presence of an abdominal mass, obstruction, or intussusception [1,8]. These non specific symptoms often overlap with other conditions, making diagnosis challenging. Differential diagnosis include volvulus, intussusception, mesenteric cysts, omental cysts, pancreatic pseudocysts, choledochal cysts, and infantile hypertrophic pyloric stenosis [4,13].

Imaging plays a pivotal role in the evaluation of abdominal masses suspected to be gastrointestinal duplications. Ultrasonography (USG) is the preferred initial diagnostic modality, as it can delineate the nature and location of the mass [14]. CT scans provide superior visualisation of the anatomical relationship between the cyst and surrounding structures, aiding in surgical planning [10]. Additional diagnostic tools, such as MRI and endoscopic ultrasonography, may also be utilised in complex cases. However, definitive diagnosis is achieved only after surgical excision and histopathological examination of the lesion. Histopathology not only confirms the diagnosis but also rules out potential malignancy, which, although rare, remains a possibility in these anomalies and is mostly reported in adults [15,16].

Surgical intervention is the treatment of choice for symptomatic duplication cysts [13]. In a case series study by Sharma S et al., six paediatrics patients diagnosed with enteric duplication cysts at various gastrointestinal locations were analysed for clinico-pathological features. Surgical intervention was performed in all cases involving cyst excision, with some requiring additional bowel segment resection and anastomosis [1]. Similarly, Tiwari C et al., also concluded that surgical intervention is the mainstay of treatment in these cases [12]. The decision-making process for asymptomatic cases is more nuanced, as management remains controversial. Some advocate for observation due to the potential risks of surgery, while others recommend prophylactic excision to mitigate the risk of complications and malignant transformation [17]. Given the variability in presentation and management considerations, an individualised approach based on the clinical scenario is essential.

CONCLUSION(S)

Gastrointestinal duplication cysts, though rare, require a definitive diagnosis due to their potential to present with varied clinical manifestations and lead to severe complications such as obstruction, perforation, haemorrhage, or malignancy. The present case highlights the importance of a multidisciplinary approach, where radiological imaging aids in localisation and characterisation, intraoperative findings provide critical anatomical insights, and histopathological evaluation confirms the diagnosis. Timely surgical intervention remains the cornerstone of management, ensuring the prevention of life-threatening complications and improving patient outcomes.

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