

Case Report

A rare case presentation of combined enteric duplication cyst both tubular and cystic

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ABSTRACT

Enteric duplication cyst are rare congenital malformations of children and can develop anywhere along the gastrointestinal (GI) tract being ileum the most frequent location, they don't communicate with GI lumen. They are usually detected prenatally or in first years of life. The size, location, type, mucosal pattern produces varied clinical presentation and different imaging findings. Ultrasonography (USG) is the most commonly used imaging method for diagnosis. A 4-year-old boy was admitted in our hospital for recurrent on and off abdominal pain, on getting USG he has diagnosed has having tubular enteric duplication cyst on both USG and computed tomography (CT) scan. Surgery performed and resection and anastomosis done. Macroscopic and histologic findings confirmed the diagnosis of enteric duplication cyst arising from ileum. Although intestinal duplications are considered to be benign lesions, they may result in significant morbidity and mortality if left untreated.

Keywords: Enteric duplication cyst, Combined cyst

INTRODUCTION

Enteric duplication cyst are rare congenital anomalies found anywhere along gastrointestinal tract (GI) tract from mouth to rectum, most commonly in ileum [33%], esophagus [20%], colon [13%], jejunum [10%], stomach [7%] and duodenum [5%].¹⁻⁴ Incidence is 1:4500 births, found in 0.2% of all children with slight male predominance.^{3,5-7}

Enteric duplication cyst is believed to develop between 4th to 8th week of embryonic development, etiology is still unknown. Several theories have been proposed to explain their pathophysiology, but no single hypothesis can justify all duplication, location and associated anomalies. The different theories lead to think that origin of enteric duplication cyst can be multifactorial.^{1,2,4,8,9} Most of the times it is associated with spinal defect, cardiac or urinary malformations. Therefore, once enteric duplication cyst is found, a search for another anomaly is needed.

Enteric duplication cyst must have three characteristics i.e., an epithelial lining containing mucosa of alimentary tract, an envelope of smooth muscle and it is closely attached to adjacent bowel by sharing common wall. Ectopic gastric mucosa is found in 20-30% of cyst, more frequently in esophagus and small bowel duplication.^{10,11}

CASE REPORT

A 4-year-old boy presented with recurrent on and off abdominal pain without any signs of obstruction, examination doesn't reveal any palpable lump. Bowel sounds are normal. An abdominal ultrasound scan showed tubular elongated cystic lesion of size 80×16 mm in left iliac region with double wall sign and inseparable from adjacent small bowel loop. A computed tomography (CT) scan confirmed the presence of thick-walled cystic lesion of 23×67 mm in left lumbar region which shows peripheral enhancement. On interval scan, lesion shows change in position. Then we planned for laparotomy and

intraoperatively found a cystic swelling which is sharing its wall with adjacent ileum, resection and anastomosis done. Postoperative period was uneventful. Microscopic findings confirmed the diagnosis of enteric duplication cyst arising from ileum with normal intestinal epithelium. Patient is discharged on 4th day without any complication.

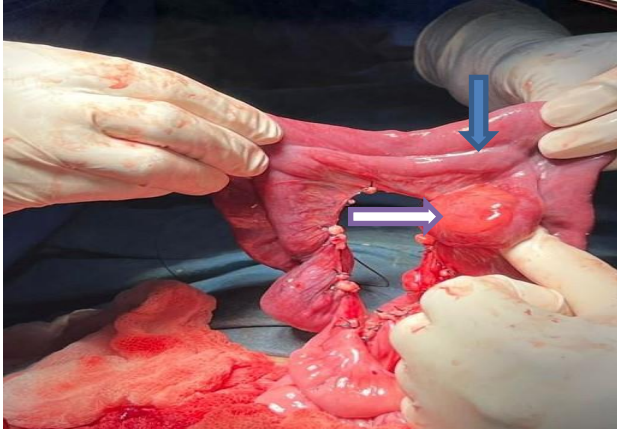


Figure 1: Intraoperative image showing both tubular (blue arrow) and cystic (yellow arrow).

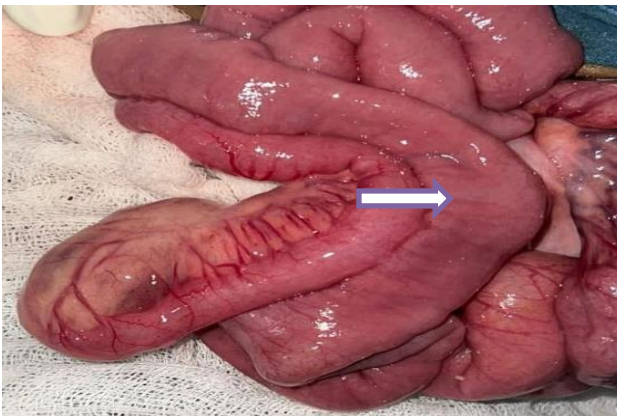


Figure 2: Intraoperative image showing both tubular variety only (white arrow).

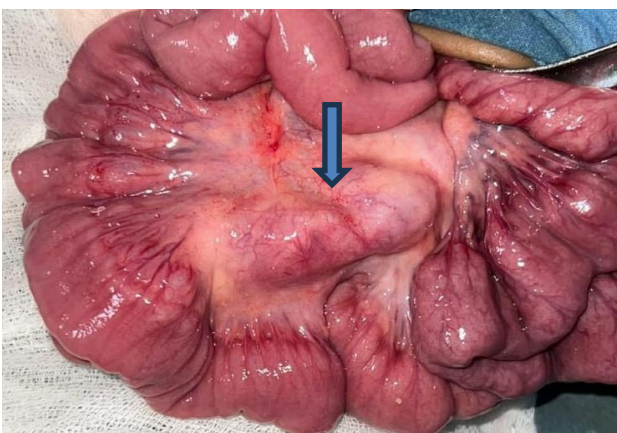


Figure 3: Intraoperative image showing cystic variety (blue arrow).

DISCUSSION

Congenital alimentary tract malformation is rare developmental mass that can occur anywhere in entirety of gastrointestinal tract. These lesions have been assigned several different names, including enterocystomas, enterogenous cyst, supernumerary accessory organs, ileum duplex.¹² Duplication of ileum represent most common form of alimentary tract duplication and typically appear as non-communicating cyst of varying size, when they take the form of long tubular segments, they tend to distend and cause extrinsic compression and thus obstructive symptoms. Because of their common location at mesenteric border, they are commonly mistaken for omental cyst which is confirmed by identification of mucosal rather than endothelial lining in omental cyst.

Abdominal cavity duplication cyst carries majority (75%) incidence among all duplication cyst in body followed by intrathoracic (20%), thoracoabdominal (5%). Jejunal and ileal carries 53% followed by colonic 13%, gastric 7%, duodenal 6%, rectal 4%. Enteric duplication cyst is characterized by epithelial lining of GI mucosa, presence of well-developed smooth muscle in wall, sharing its wall with adjacent bowel. Enteric duplication cyst is presented with pain and vomiting usually, sometimes it may present as bleeding, perforation from ulcer, gastric outlet obstruction in foregut duplication cyst, intussusception in midgut duplication cyst, it may range from obstruction to incontinence in hindgut duplication cyst.

Approximately 2/3rd of all intestinal duplication are discovered within first 2 years of life with 1/3rd identified in newborn period.¹² They are slightly more common in males, etiopathogenesis is explained by 4 theories named partial twinning theory, split notochord theory, canalization defects theory, environmental factors theory.¹² Li characterized these cysts into type-I – parallel type, in which one vessel perfusing the normal bowel and other perfusing duplication cyst, type- II- intramesentric type, where vessel traverse the duplication on its way to perfusing the native intestine.¹²

Enteric duplication cyst generally identified in antenatal ultrasonography (USG) and post-natal USG in case of both thoracic and abdominal variety, X-ray is to look for the intussusception, volvulus, CT scan delineate the anatomy and to find synchronous lesion, magnetic resonance imaging (MRI) is necessary if neurologic symptoms of spinal cord compression observed, Tc99m pertechnetate scan to detect ectopic gastric mucosa.

CONCLUSION

These cysts don't require any intervention if it is asymptomatic, but it is better to remove as soon as possible since there is a possibility of malignant transformation but it is rare. Pre-operative antibiotics and bowel preparation is done and cyst is removed completely along with attached bowel as both share common blood supply and

the anastomosis is done. Alternative approach includes marsupialization of cyst; this consist of partial cyst excision with mucosal stripping of remaining cyst wall to preserve normal anatomy. Postoperative complications include bleeding, infection and bowel obstruction.

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Ethical approval: Not required

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