Coc Cer Derg/Turkish J Ped Surg 2022;36(1): 70-73
doi: 10.29228/JTAPS. 53679

Case Report

Çocuk Cerrahisi Dergisi
Turkish Journal of Pediatric Surgery
Volume 36 Number 1 2022

A Rare Cause of Cecal Obstruction: Enteric duplication cyst

Ahmet Hikmet Şahin

Balıkesir University, Department of Pediatric Surgery, Balıkesir-Türkiye

ABSTRACT

Although it can be seen at any age, enteric duplication cysts are seen in 80% of the first two years of life, especially in the first three months. 33-53% of enteric duplications are diagnosed in the ileum. In this article, it is aimed to discuss the situation in the literature due to two cases who presented with cecal obstruction and were diagnosed with enteric duplication.

First case; Gas density was observed in the left upper quadrant on the direct abdominal X-ray taken in a two-year-old male patient who presented with vomiting. In the abdominal ultrasonography, a cystic structure with 3 cm diameter septa was detected in the subhepatic region.

The second case: a 14-month-old female patient, complaining of vomiting and restlessness, had gas density in the left upper quadrant on the abdominal X-ray and a cystic mass in the right lower quadrant on the abdominal ultrasonography.

Enteric duplication cysts, which also cause life threatening, are rare. Accurate follow-up of pregnancies is thought to provide early prenatal detection of enteric duplication cysts and prevent possible complications.

Keywords: *Caecum, surgery, intestinal duplication cyst, obstruction, child*

Geliş/Received: 30.04.2021
Kabul/Accepted: 10.06.2021
Published online: 01.04.2022

Cite as: Şahin AH. A Rare Cause of Cecal Obstruction: Enteric duplication cyst. Coc Cer Derg/Turkish J Ped Surg 2022;36(1): 70-73. doi: 10.29228/JTAPS. 53679

Ahmet Hikmet Şahin
Balıkesir University, Department of Pediatric Surgery, Balıkesir-Türkiye
drhikmetsahin@gmail.com
ORCID: 0000-0002-9134-9800

Introduction

Gastroenteric duplications occur in 1/4500 births and predominantly in males [1,2]. Although it is seen at any age, 80% of them are seen in the first two years of life, especially in the first three months. The most common site of duplications is the ileum with a rate of 33-53% [2,3]. The mechanism by which enteric duplication occurs is not entirely known. Split Notokord Theory, Luminal Recanalization Theory, Persistent Embryonic Diverticulum Theory and Intrauterine Vascular Accident Theory are the theories mentioned in understanding the mechanism by which enteric duplication occurs. Even proposing different theories is an indication that the enteric duplication is multifactorial[2].

Enteric duplications cysts can be single or multiple, spherical or tubular, isolated or connected[4]. The spectrum of symptoms is broad in enteric duplication.

In some cases, it is not causing symptoms, but in some cases may have life-threatening symptoms such as volvulus, gastrointestinal bleeding, perforation, and invagination. Prenatal and postnatal USG, abdominal CT, and MRI imaging are used in diagnosis. Enteric duplication cysts can cause significant morbidity and mortality, for reasons such as ulceration of the intestinal mucosa, massive bleeding or intestinal perforation, if left untreated. Surgical treatment is recommended in all cases [3].

This paper will present literature information due to two cases presenting with cecal obstruction and found to have enteric duplication.

Case 1:

A 2-year-old male patient weights 9 kg. He applied to the emergency department with the complaint of vomiting. In his anamnesis, stated that he had been unable to poop for a day and had bilious vomiting. There is no information about intestinal anomaly in the antenatal screening. In his physical examination, it was determined that there was mass in the epigastric region, and the skin was pale and dry.

On standing direct abdominal radiography, intestinal loops were stuck in the left upper quadrant, and the pre-diagnosis of ileus was admitted to the intensive care unit. Intravenous fluid replacement and prophylactic antibiotics (cefazolin 50 mg/kg) were administered to the patient. In the abdominal ultrasonography performed during the follow-up, a 3 cm diameter cystic structure with septa (possibly intestinal volvulus or omental tortion) in the subhepatic area was detected. Abdominal exploration was performed after informing the parents. The abdomen was entered with a transverse incision fitting in the right lower quadrant. It was observed that the ileal segments were distended, and the cecum was obstructed with a firm mass in the abdomen.

When the mass was eluxated from the incision, enteric duplication cyst (EDC) was detected in the mesenteric area. It was observed that the enteric duplication cyst completely closes the ileocecal passage. The serosa of the cyst is incised, and the cyst was removed from the cecum and mesentery by dissected (Figure 1).

Serosa was closed with 4/0 vicryl with one by one suture. Ileocecal transition was observed to be expected. Appendectomy was also applied to the patient. The patient was fed with aqueous food on the first postoperative day and was discharged on the second postoperative day. Pathological examination confirmed an enteric duplication cyst, and no malignancy was detected in the material.

Case 2:

A 14-month-old female patient weighs 8 kg. She applied to the emergency department with the complaint of vomiting and uneasiness. In her anamnesis, it was learned that bilious vomiting started 3-4 hours ago, and there was no known disease. There is no information about intestinal anomaly in the antenatal screening. On standing direct abdominal radiography, it was observed that the intestinal loops were squeezed into the left upper quadrant. The patient was admitted to the pediatric intensive care unit with the pre-diagnosis of ileus and intravenous fluid replacement, and a prophylactic antibiotic (cefazolin 50 mg/kg) was administered. In the abdominal ultrasonography conducted during the follow-up, a cystic mass was detected in the right lower quadrant. After informing the parents, the exploration decision was made. In exploration, it was found that a cystic mass completely closed the ileocecal passage, and the ileal segments were distended. It was determined that the cyst was too attached to the cecum to be dissected from the cecum. The ileocecal area was resected together with the enteric duplication cyst (Figure 2).

The ileocolic end lateral anastomosis was sutured in two layers with 4/0 vicryl. The patient started to be fed with aqueous food on the third postoperative day and was discharged on the fourth postoperative day. Pathological examination of the resected material confirmed an enteric duplication cyst, and no malignancy was detected in the material.

Discussion

Although there are different theories regarding the development of enteric duplication cysts, it is thought to develop between the 4th and 8th week of embryological life, often due to errors during notochord separation [5]. Duplication cysts lined with gastrointestinal epithelium are located on the mesenteric side of the gastrointestinal tract. Enteric duplication cysts that cause acute or chronic complaints such as invagination, obstruction, bleeding, perforation, and malignant transformation are usually detected incidentally [6,7].

Prenatal ultrasonography may show an intra-abdominal mass in the 2nd or 3rd trimester of pregnancy in 20% to 30% of cases[8]. It may be confused with Meckel diverticulum or invaginations [3]. Double-wall image and the presence of peristalsis in ultrasonography are determinative for enteric duplication cysts. Postnatal USG, contrast-enhanced computed tomography, abdominal magnetic resonance imaging can distinguish them from other intra-abdominal cystic lesions[9].

Figure 1. Direct abdominal radiography and operation view

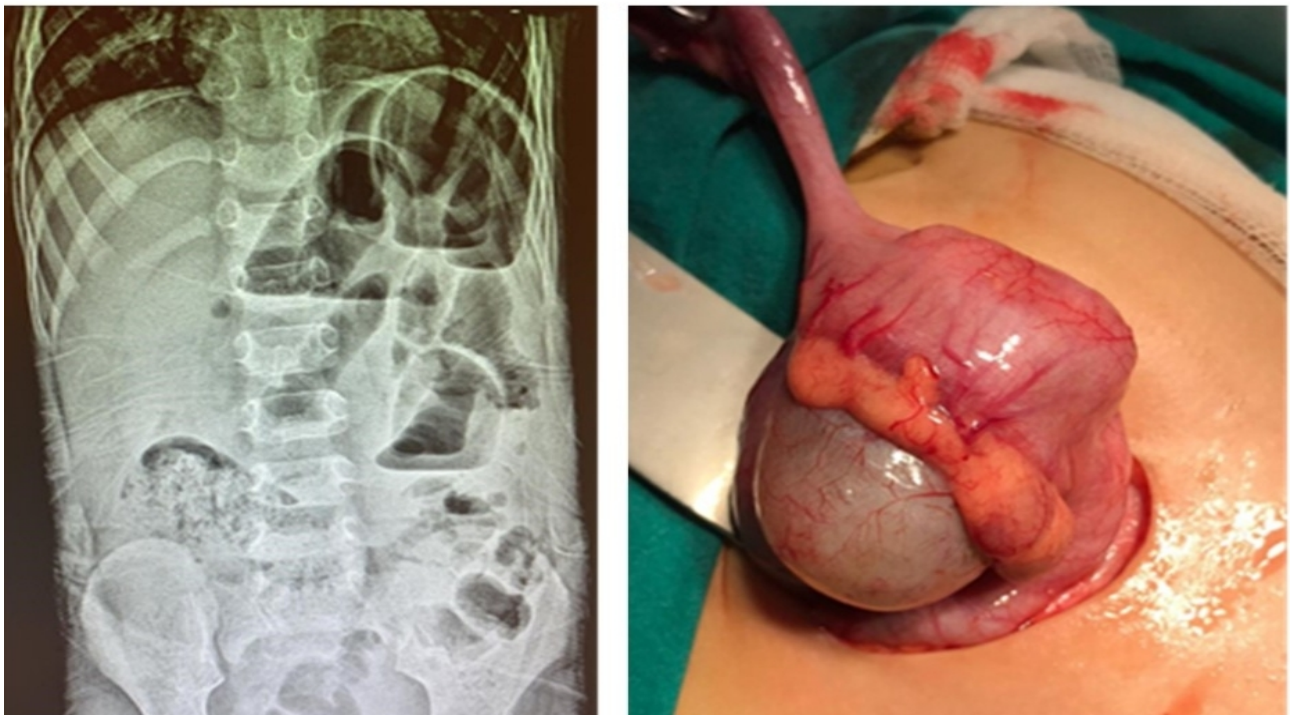
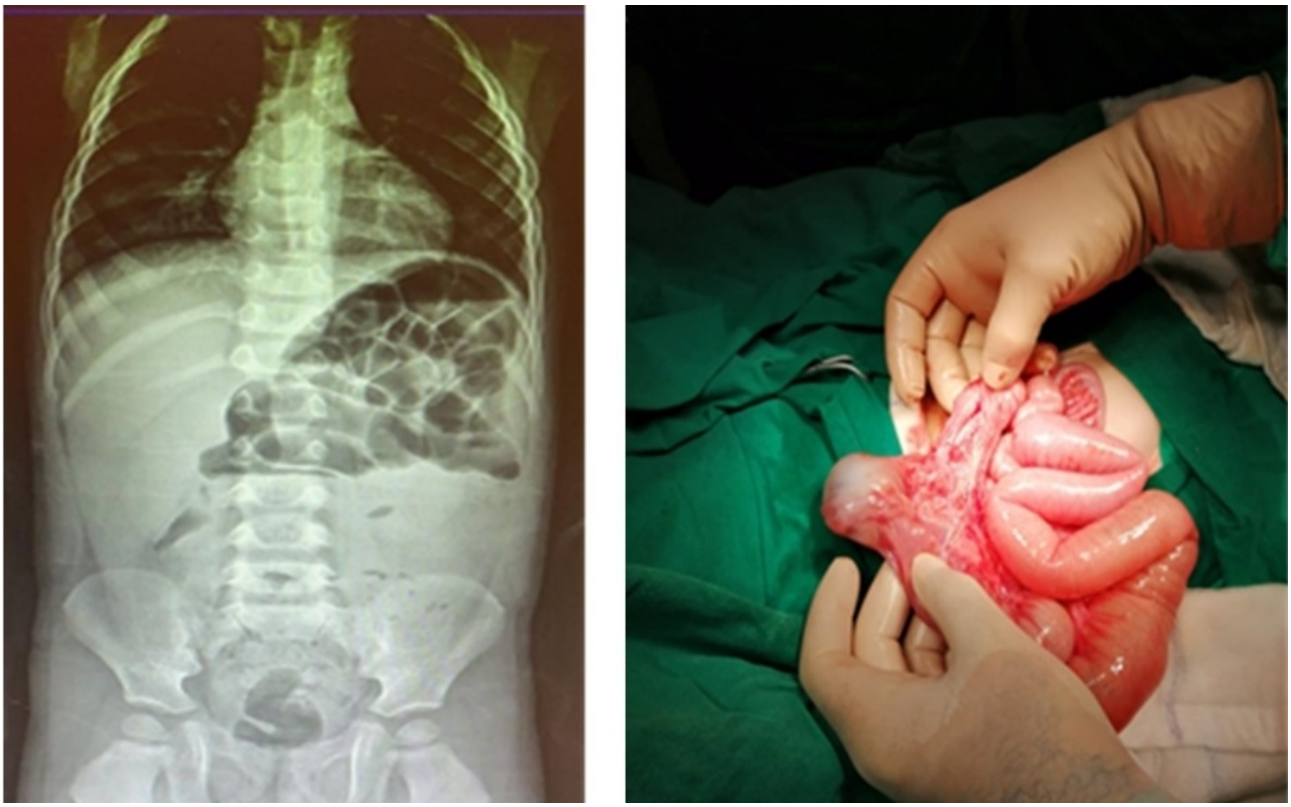


Figure 2. Direct abdomen radiography and surgical finding



There is no consensus about the postnatal fate of enteric duplication cysts detected during the prenatal period in the literature. The manner of intervention in enteric duplication cysts detected during the prenatal period and without any complaints is not clear [10,11]. Nevertheless, when there is an acute-life-threatening condition such as volvulus, invagination, or perforation, surgical intervention is applied. In cases detected during the prenatal period, according to Fahy et al., it was reported that 41% were operated on due to early complaints, 59% had protective resection during the asymptomatic period[10]. Enucleation may be sufficient depending on the location and size of enteric duplication cysts, but segmental resection of the affected organ is required in most cases[11]. In some cases, EDC is removed laparoscopically [4].

Both cases presented in this article were brought to the emergency department with the complaint of vomiting, which is a sign of intestinal obstruction. In the abdominal USG performed, enteric duplication cysts in the ileocecal region were found in accordance [2,3] with the literature. Enteric duplication cyst was detected in both cases in the postnatal period, and surgical intervention was required. Parents of both cases are not relatives, their mothers were not followed up during their pregnancy, and prenatal USG was not performed. Their mothers stated that they did not experience any environmental exposure that could cause congenital anomalies during pregnancy. Both cases are isolated enteric duplication cysts. There are no other congenital anomalies. When enteric duplication cyst resection was performed in the first case, in the second case, ileocecal resection was performed with the cyst. This is important in terms of showing that each case should be evaluated separately. No finding of malignancy was detected in the pathological examination of the resection materials of the cases. The cases were discharged with complete recovery.

Conclusion

Although enteric duplication cysts are rare, the symptoms they cause can be life-threatening for the cases. To minimize the danger, enteric duplication cysts can be detected by performing prenatal follow-up of mothers during pregnancy and strict follow-up of the detected cases postnatal period. Therefore, prenatal period pregnancy and strict follow-up and postnatal baby follow-up should not be neglected. It should be considered that the location of enteric duplication cysts may change, and when detected, surgical treatment should be planned according to the patient.

Declaration of Conflicting Interests

The author declared no potential conflicts of interest concerning the research, authorship, and publication of this article.

Funding

The author(s) received no financial support for the research, authorship, and publication of this article.

Ethical Approval/Patient Consent

Written consent from the patients' families and ethical approval from the relevant institution was obtained.

References

1. J. L. Grosfeld, J. A. O'Neill, A. G. Goran, E. W. Fonkalsrud and A. A. Caldamone, Pediatric Surgery - 2-Volume Set, Mosby, 2006, pp. 1389-1398.
2. C. S. Nebot, R. L. Salvador, E. C. Palacios, S. P. Aliaga, and V. I. Pradas, "Enteric duplication cysts inchildren: varied presentations, varied imaging findings," Insights Imaging, vol. 9, no. 6, pp. 1097-1106., 2018.
3. S. Beura, B. N. Mishra, R. K. Joshi, and P. P. Pahi, "Enteric duplication cyst in a neonate: A case report," International Journal of Surgery Science, vol. 4, no. 2, pp. 303-305., 2020.
4. S. K. Thompson, A. L. Wong, C. L. Trevenen and J. H. MacGregor, "Enteric duplication cyst," Am J Surg, vol. 187, no. 2, pp. 316-8., 2004.
5. D. Ayadi, A. Bezzine, E. B. Hamida and Z. Marrakchi, "Fetal Cyst Reveling Retroperitoneal Enteric Duplication," J Indian Assoc Pediatr Surg, vol. 22, no. 1, pp. 60-61, 2017.
6. L. M. v. Zitteren, M. Ruppert, B. O. d. Beeck and M. Wojciechowski, "Infected enteric duplication cyst," BMJ Case Rep, vol. 21, pp. 1-2, 2017.
7. Y. Korotkaya, E. Dunn, A. Aybar, J. Crino, S. Alaish, and A. Scheimann, "Cecal Duplication Presenting With Hematochezia in an Infant," Clinical Pediatrics, vol. 58, no. 11-12, p. 1338–1340, 2019.
8. S. Iijima, "A Wandering Abdominal Mass in a Neonate: An Enteric Duplication Cyst Mimicking an Ovarian Cyst," Case Reports in Pediatrics, 2017.
9. Neazy, A. Almatrfi, W. Alharbi, and A. Abokrecha, "Two different presentations of intestinal duplication cyst in pediatric age group," Saudi Surg J, vol. 6, pp. 100-3., 2018.
10. S. Fahy and A. Pierro, "A Systematic Review of Prenatally Diagnosed Intra-abdominal Enteric Duplication Cysts," Eur J Pediatr Surg, vol. 29, no. 1, pp. 68-74, 2019.
11. A.-R. L. Olajide, A. A. Yisau, N. A. Abdulraseed, I. O. Kashim, A. J. Olaniyi and A. O. Morohunfade, "Gastrointestinal duplications: Experience in seven children and a review of the literature," Saudi J Gastroenterol, vol. 16, pp. 105-9, 2010.