Erciyes Med J 2020; 42(3): 336–8 • DOI: 10.14744/etd.2020.43402
CASE REPORT – OPEN ACCESS

Gastric Duplication Cyst in an Infant Presenting with Melena

Derya Altay[1], Süreyya Burcu Görkem[2], Kemal Deniz[3], Kerametin Uğur Özkan[4], Duran Arslan[1]

ABSTRACT

Background: Gastrointestinal duplication cysts are rare congenital anomalies that are most often localized in the ileum. The condition is usually diagnosed within the first year of life.

Case Report: Here, we report an infantile case presented with severe anemia and melena accompanied by a gastric duplication cyst.

Conclusion: Duplication cysts should be kept in mind in the differential diagnosis of melena.

Keywords: Duplication cyst, gastric, infant, melena

Cite this article as:
Altay D, Görkem SB, Deniz K, Özkan KU, Arslan D. Gastric Duplication Cyst in an Infant Presenting with Melena. Erciyes Med J 2020; 42(3): 336-8.

[1]Department of Pediatric Gastroenterology, Hepatology and Nutrition, Erciyes University Faculty of Medicine, Kayseri, Turkey
[2]Department of Pediatric Radiology, Erciyes University Faculty of Medicine, Kayseri, Turkey
[3]Department of Pathology, Erciyes University Faculty of Medicine, Kayseri, Turkey
[4]Department of Pediatric Surgery, Erciyes University Faculty of Medicine, Kayseri, Turkey

Submitted
29.01.2020

Accepted
02.03.2020

Available Online Date
15.06.2020

Correspondence
Derya Altay,
Erciyes University Faculty of Medicine, Department of Pediatric Gastroenterology, Hepatology and Nutrition, Kayseri, Turkey
Phone: +90 352 207 66 66
e-mail:
dr.deryaaltay@gmail.com

INTRODUCTION

Duplication cysts, one of the rare congenital anomalies of the gastrointestinal tract, may occur at any location from the mouth to the anus. The condition is identified in approximately one out of 4500 live births and is most commonly located in the ileum; however, gastric duplication cysts are rarely seen (1). Duplication literally means "copy", it is adjacent to the gastrointestinal tract at its localization, contains a thick, smooth muscle layer, and is covered by the digestive tract epithelium (2). There may be different clinical findings according to its location, such as vomiting, abdominal pain, bleeding, and distension. Cysts may be symptomatic or incidentally detected during surgery, and they are treated surgically by cyst excision. Here, we report on an infantile patient who presented with severe anemia and melena and was accompanied by a gastric duplication cyst.

CASE REPORT

A three-month-old male infant was admitted to the Pediatric Emergency Department due to a case of black stools. From his medical history, it was gathered that, on the first day of his life, he was operated on due to suspicion of intestinal perforation, although no pathology was detected at the operation. Upon the patient's physical examination, his weight was 3190 g, height was 54 cm, head circumference was 37 cm, the skin was markedly pale, and there was an incision scar on the abdominal midline. The patient was hospitalized due to a hemoglobin value of 4 g/dL and hemodynamic instability, which were identified through laboratory tests. After the patient had received erythrocyte suspension, his control hemoglobin value was 9.5 g/dL. Posteroanterior chest radiograph of the patient was normal (Fig. 1a). Esophagogastroduodenoscopy was performed to investigate the etiology of upper gastrointestinal bleeding, and no pathology was observed during endoscopy. Our patient was discharged for outpatient follow-up. Three weeks post-discharge, he was brought back due to a case of black stool, and this time, his hemoglobin level was 2.7 g/dL. After two erythrocyte suspensions were applied, the patient's control hemoglobin level increased to 7.3 g/dL. Gastrointestinal bleeding scintigraphy was performed on the patient, who exhibited no significant pathology in abdominal ultrasonography, and no bleeding focus was detected through scintigraphy. This result was associated with the patient's lack of active bleeding during scintigraphy. Subsequently, computerized tomography of the upper and lower abdomen was performed. In the thorax sections of computed tomography, a cystic, space-occupying formation of approximately 7x3 cm was observed located in the anterior of the spleen and the posterior of the stomach in the upper abdomen near the second-third part of the duodenum, extending to the right side of the esophagus. The radiologist believed the formation could be an enteric duplication cyst exhibiting herniation to the thorax. Magnetic resonance imaging of the thorax and abdomen was performed, and the result was consistent with an enteric duplication cyst extending to the esophagus (Fig. 1b, Fig. 2). No contrasting material transition to the suspicious duplication cyst was identified through barium contrast graphy, and the treitz ligament was fixated on the right side of the vertebral column. The patient was operated on by the Pediatric Surgery Department. The right posterolateral thoracotomy was performed and the duplication cyst localized in the gastric cardia, extending into the esophagus, was excised. In

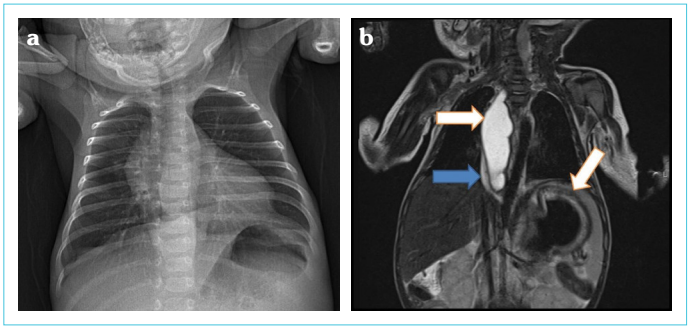

Figure 1. (a) Anteroposterior chest radiograph of the infant demonstrates no abnormality. (b) Coronal T2 HASTE image through thorax and upper abdomen demostrates duplication of esophagus and stomach (white arrows). Normal anatomical esophagus is shown (blue arrow)

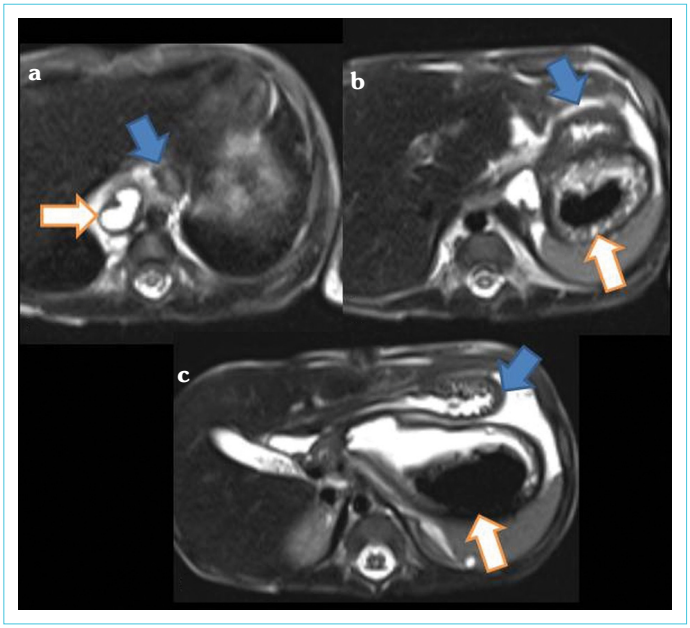

Figure 2. Consecutive axial T2 HASTE images through upper abdomen show duplication of esophagus (a) and stomach (b, c) (white arrows). Normal anatomical esophagus and stomach are shown as blue arrows (a–c)

histopathological evaluation, gastric duplication cyst was contained gastric tissue and the excised duplication cyst also showed mature cartilage islets in the submucosa of the cyst wall, but the ciliary epithelium of the respiratory tract was not observed (Fig. 3). Two days after the gastric duplication cyst was excised, the patient was fed orally. Enteral feeding of the patient was gradually increased, and the patient's general condition was good; clinical follow-up remains ongoing. The patient's consent was obtained for this study.

DISCUSSION

Duplication cysts are usually diagnosed within the first year of life. Although the exact cause of the duplication cysts' formation is unknown, hypotheses, such as pause, partial twinning, and split notochord in the recanalization phase of the primitive intestine during the embryologic period, have been proposed (3).

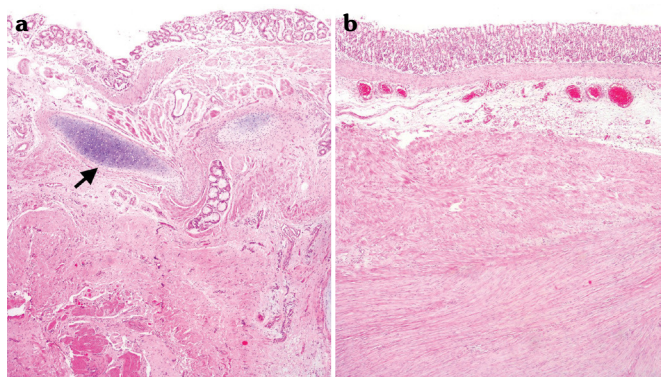

Figure 3. (a) Figure of cyst wall with mature cartilage (arrow) in submucosa and (b) figure of the gastric wall containing normal layers of tissue (hematoxylin eosin; x40)

Gastric duplication cysts constitute 4–9% of the intestinal duplication cysts and are not associated with the gastric lumen (2). Patients with gastric duplication cyst may be asymptomatic or present with abdominal pain, vomiting, weight loss, gastrointestinal bleeding, and gastric outlet obstruction (4, 5). In our case, the patient was admitted to the hospital several times due to complaints of melena as a result of upper gastrointestinal bleeding. Although the patient's upper gastrointestinal endoscopy was normal, it was possible to diagnose a duplication cyst using advanced imaging methods. Gastric duplication cysts can be diagnosed in the infantile period or may remain silent until adulthood. However, the majority of gastric duplication cysts are diagnosed in the infantile period (6).

Barium graphy, ultrasonography, endoscopic ultrasonography, computed tomography, and magnetic resonance imaging are valuable for the diagnosis of duplication cysts. Jehangir et al. (7) evaluated the cases with 35 enteric duplications and emphasized that they observed small bowel duplication most frequently; the duplication cysts were more common in males than in females. The authors also stated that 55% sensitivity during ultrasonography for diagnosis, along with high clinical suspicion, could increase the diagnostic value of ultrasonography. Sonographically, a double-wall view of the hypoechogenic outer muscular layer over the echogenic mucosal layer is typical for duplication cysts. In our case, that the cysts were not detected in the initially performed abdominal ultrasonography was explained by the prolongation of the cyst toward the thorax and the failure to issue a clinical diagnosis. The diagnosis and detection of the duplication cysts were performed using computerized tomography. Generally, abdominal ultrasonography is the first step in researching the etiology of bleeding; for all that computed tomography and magnetic resonance imaging should not be delayed if no pathology is detected using ultrasonography.

Histopathological evaluation is the definitive way to confirm the diagnosis. Histopathological examination of the patient's duplication cyst was revealed as gastric tissue, and interestingly, mature cartilage islets were included in the submucosa of the cyst wall. The ciliary epithelium of the respiratory tract was not observed, and it was interpreted as developmental ectopia of cartilage tissue in the submucosa. Otherwise, gastric duplication cysts originating from respiratory diverticulum have been reported by Khoury and Rivera (8). Because there was no ciliary epithelium in our case, the duplication cyst was not evaluated in relation to the respiratory tract.

The treatment of duplication cysts in symptomatic patients is surgical cyst excision. However, asymptomatic patients can be followed by imaging methods and can be operated on if the cysts grow or become symptomatic (2). In our case, cyst excision was performed for the gastric duplication cyst extending to the esophagus, and thus gastrointestinal bleeding of the patient was stopped.

CONCLUSION

Gastric duplication was the cause of upper gastrointestinal bleeding in our patient. The consideration of duplication cysts along the differential diagnoses of gastrointestinal bleeding within the first months of life is significant for the reduction of morbidity and mortality. It is especially crucial that pediatricians are aware of duplication cysts and should work closely with the radiology department to diagnose early. Interestingly, in this case, gastric duplication cyst also showed mature cartilage islets in the submucosa of the cyst wall.

Informed Consent: Written informed consent was obtained from patients who participated in this study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept – DA, SBG; Design – DA; Supervision – DA, DuA; Resource – DA, SBG, KD; Materials – DA, SBG, KD, KUO; Data Collection and/or Processing – DA, SBG; Analysis and/or Interpretation – DA, KD; Literature Search – DA; Writing – DA; Critical Reviews – DuA.

Conflict of Interest: The authors have no conflict of interest to declare.

Financial Disclosure: The authors declared that this study has received no financial support.

REFERENCES

1. Fang Y, Gao T, Yang H, Ma S, Li Q, Zhou PH. Removal of an infant's gastric duplication cyst through endoscopic submucosal dissection: A case report. Medicine (Baltimore) 2019; 98(12): e14820. [CrossRef]
2. Liu R, Adler DG. Duplication cysts: Diagnosis, management, and the role of endoscopic ultrasound. Endosc Ultrasound 2014; 3(3): 152–60. [CrossRef]
3. Sharma S, Yadav AK, Mandal AK, Zaheer S, Yadav DK, Samie A. Enteric Duplication Cysts in Children: A Clinicopathological Dilemma. J Clin Diagn Res 2015; 9(8): EC08–11. [CrossRef]
4. Youssef A, Ibrahim A, Al Shehabi Z, Omran A, Sharara AI. Gastric Duplication Cyst Presenting as Massive Gastrointestinal Bleeding. Pediatr Gastroenterol Hepatol Nutr 2019; 22(2): 189–92. [CrossRef]
5. Khan Khattak MA, Nisar MU, Sikander S, Akthar N. Gastric Duplication Cyst: A Rare Congenital Anomaly Causing Gastric Outlet Obstruction. J Ayub Med Coll Abbottabad 2019; 31(3): 466–8.
6. Singh JP, Rajdeo H, Bhuta K, Savino JA. Gastric duplication cyst: two case reports and review of the literature. Case Rep Surg 2013; 2013: 605059. [CrossRef]
7. Jehangir S, Ninan PJ, Jacob TJ, Eapen A, Mathai J, Thomas RJ, et al. Enteric duplication in children: Experience from a tertiary center in South India. J Indian Assoc Pediatr Surg 2015; 20(4): 174–8. [CrossRef]
8. Khoury T, Rivera L. Foregut duplication cysts: a report of two cases with emphasis on embryogenesis. World J Gastroenterol 2011; 17(1): 130–4. [CrossRef]