A Different Treatment Modality for a Caecal Intramural Duplication Cyst in an Infant: Case Report

Bir Bebekte Çekal İntramural Duplikasyon Kisti İçin Farklı Bir Tedavi Yaklaşımı

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ABSTRACT Duplication cysts are uncommon congenital abnormalities that may occur at all levels of the gastrointestinal tract, from mouth to anus. In most cases, end-to-end anastomosis after segmental resection of a bowel with duplication cysts is the preferred choice of treatment. A 7-month-old female presented with occasional vomiting episodes since birth. Previous medical examinations had revealed no abnormality. She underwent surgical intervention due to acute abdomen with an eventual diagnosis of duplication cyst. In our case, both the caecum and ileocaecal valve were preserved. She was treated with partial excision and removal of the mucosal lining, which is an uncommon treatment approach and the result was satisfactory. We suggest that removal of the caecum and ileocaecal valve along with the cyst is a treatment option; however, alternative treatment options should also be considered.

Key Words: Digestive system; infant; intussusception

ÖZET Duplikasyon kistleri gastrointestinal sistemde ağızdan anüse kadar tüm seviyelerde ortaya çıkabilen nadir doğumsal anomalilerdir. Genellikle olguların çoğunda total eksizyon önerilir. Eksizyon sırasında çekum ve ileoçekal valv de çıkarılır. Doğumdan itibaren sürekli kusma atakları yakınması ile başvuran 7 aylık kız olgu akut batın nedeni ile opere edildi ve çekal intramural duplikasyon kisti tanısı aldı. Parsiyel eksizyon ve mukoza soyma işlemleri ile operasyon esnasında hem çekum hem de ileoçekal valv korundu. Sonuç tatmin edici bulundu. Farklı bir tedavi yaklaşımı kullanıldığı için bu olgunun bildirilmeye değer olduğu düşünüldü. Sonuç olarak, kist ile beraber çekum ve ileoçekal valvin çıkarılması, kullanılan bir tedavi yöntemidir. Buna ilaveten alternatif tedavi seçeneklerinin de düşünülmesi gerekir.

Anahtar Kelimeler: Sindirim sistemi; bebek; intüsüsepsiyon

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Duplication cysts are uncommon congenital abnormalities first identified by Ladd.[1] They may occur at all levels of the gastrointestinal tract, from mouth to anus.[2,3] Usually, they present with a tubular or cystic structure. Typical characteristics of duplication cysts are tight attachment to the gastrointestinal tract, a well-developed muscular wall along with an internal epithelium originating from the epithelial tissue of the gastrointestinal tract, and localization on the mesenteric side.[3] Clinically, they may present with intussusception, obstruction, volvulus, malignity, hemorrhage, and rarely perforation.[4,5] Reoprts on caecal intramural duplication cysts associated with intussusception is rare. In such cases, end-to-end anas-

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tomosis after segmental resection of a bowel with duplication cysts is the preferred method of treatment. However, in our case, both the caecum and ileocaecal valve were preserved.

CASE REPORT

A 7-month-old female presented with occasional vomiting episodes since birth. Previous medical examinations had revealed no abnormality. She has had severe vomiting accompanied by diarrhea for 20 days and eventually received medical treatment on an outpatient basis. She had previously been hospitalized for one week because of persistent vomiting and diarrhea, during which time the treatment was continued. Her diarrhea had disappeared during the two days before her arrival at our clinic, but her vomit had become yellow-green in color. Her abdomen started to show distension. She was referred to our clinic with a presumptive diagnosis of intussusception.

PHYSICAL EXAMINATION

The patient's abdomen was highly distended and was not involved in respiration. Intestinal sounds were reduced and there were occasional metallic sounds. Palpation revealed tenderness in the right abdomen and rectal examination revealed blood in stool and a mobile mass.

RESULTS

The white blood cell count was 15 000/mm^3. Direct erect abdominal x-ray revealed multiple air-fluid levels. Abdominal ultrasound showed invaginated intestinal segments in the right lower quadrant.

After undergoing fluid and electrolyte replacement and giving "informed consent," the patient was prepared for the operation. Surgery was performed through a right paramedian incision. After reaching the abdomen by dissecting the cutaneous layers, a low amount of reactional and serous fluid was found and was aspirated. An invaginated intestinal segment beginning at the ileum and extending to the sigmoid colon was shown via surgical exploration and was manually reduced. Further exploration revealed that the edema had become significant over the antimesenteric area of the caecal wall and that there was a 1 cm nonencapsulated cystic mass located just underneath the serosa within the intestinal wall (Figure 1). A puncture into the mass revealed a sticky serous fluid. We failed to remove it en masse; however, the anterior portion of the cystic wall was removed and the mucosa of the posterior portion was scraped (Figure 2). The removed anterior portion of the cystic wall was placed into formaldehyde solution for examination.

HISTOPATHOLOGICAL ANALYSIS

The intestinal specimen was fixed in 4% formalin and was embedded in paraffin, and 5-7 μ sections were stained with hematoxylin and eosin (H&E) for microscopic evaluation. Each entire section was observed under magnification (x40). The structure, which was defined as an intestinal duplication cyst, was lined with simple columnar epithelium and mucous cells with the occasional presence of glands. Vessels were found to be congested as a result of the thickened muscular layer.

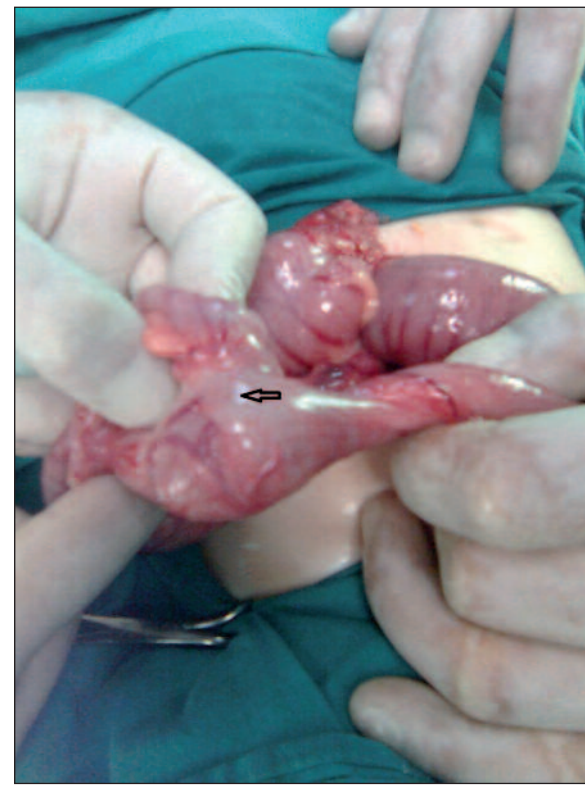

FIGURE 1: Cystic mass in the antimesenteric area of the caecal wall.
(See for colored form http://tipbilimleri.turkiyeklinikleri.com/)

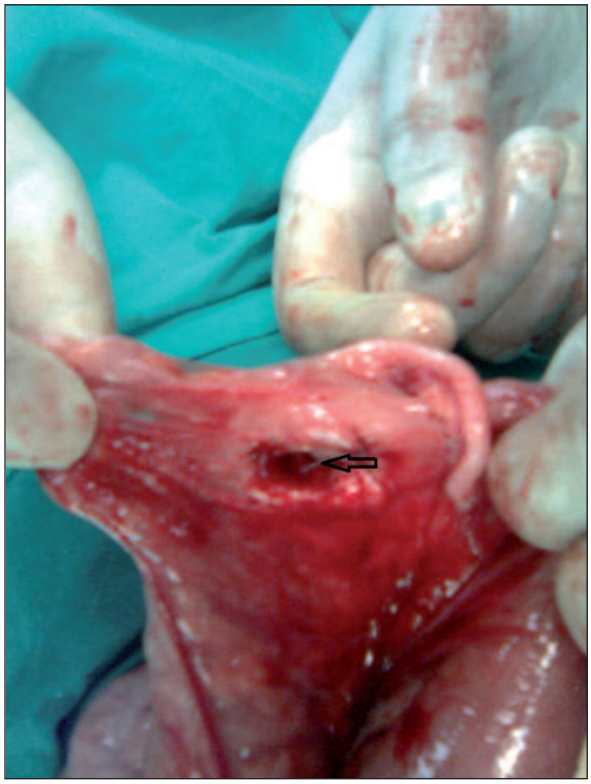

FIGURE 2: Anterior portion of the cystic wall was removed and the mucosa of the posterior portion was scraped.
(See for colored form http://tipbilimleri.turkiyeklinikleri.com/)

Oral therapy was started on the fourth postoperative day and the patient tolerated it. She was discharged on the sixth day. The 36-month follow-up revealed no complications.

DISCUSSION

While cystic duplications of the colon can be asymptomatic, they can also present with intussusception, hemorrhage, mass, or intestinal obstruction.[6,7] Some case reports involve necrosis due to intestinal volvulus. Uncommon malignancy is another complication that may develop in cases of colon duplication in patients around 35 years of age. Inoue and Nakamura studied cases of duplication cysts that had developed into malignancies with a reoprted incidence of 67% for colon and rectum duplications.[8] Duplication cysts have been reported to cause hemorrhage in cases where gastric mucosa is involved. Perforation is a less common complication. Invagination is one of the causes of acute abdominal pain among children. It often occurs for no apparent reason, beginning with intussuception of the lead point and the intestines and then advancing on. This lead point may be associated with a lymph node or luminal pathology. Caecal duplication cysts rarely result in intussusception. Usually, obstruction arises due to the mass effect.[9,10]

End-to-end anastomosis after segmental resection of the bowel with duplication cysts is the preferred choice of treatment in those cases.[11] Although duplication cysts may be clinically asymptomatic, it is recommended that they be removed by total excision due to their high rate of complications.[4,5,8]

The symptoms of intestinal duplication cysts vary according to the size, morphology, and location of the cysts. Common symptoms include acute intestinal obstruction, vomiting, recurrent abdominal pain, recurrent gastrointestinal bleeding, intussusception, constipation, or an incidental detection. Our patient with a concurrent intramural caecal duplication cyst and intussusception is an unusual case; there are only a few similar reports in the literature. She had suffered from vomiting episodes since birth. This may be attributed to the disruption of the valve mechanism and luminal motility caused by the duplication cyst, which was located close to the caecal and ileocaecal areas. Following removal of the cyst, the persistent vomiting episodes disappeared.

Removing the mucosal lining instead of total excision has been reported to produce successful results in cases of gastric duplication cysts.[12-14] Our patient was treated accordingly by partial excision and removal of the mucosal lining with a satisfactory result. The case was reported due to the alternative treatment approach.

In conclusion, we believe that removing the caecum and the ileocaecal valve along with the duplication cyst is an acceptable approach; however, alternative treatment options should also be considered. Moreover, we suggest that caecal or ileal small duplication cysts should be considered in infants with intussusception; therefore, exploration should be carefully conducted.

REFERENCES

1. Ladd WE. Duplications of the alimentary tract. South Med J 1937;30(4):363-71.
2. Bower RJ, Sieber WK, Kiesewetter WB. Alimentary tract duplications in children. Ann Surg 1978;188(5):669-74.
3. Öztekin O, Tezel G, Kalay S, Demirezen S, Oygür N. [Meconium peritonitis and nonimmun hydrops fetalis: presentation of two cases]. Turkiye Klinikleri J Pediatr 2011;20(3): 256-60.
4. Wrenn EL, Hollabaugh RS. Alimentary tract duplications. In: Ashcraft KW, ed. Pediatric Surgery. 2nd ed. New York: Saunders; 2000. p. 527-39.
5. Kuo HC, Lee HC, Shin CH, Sheu JC, Chang PY, Wang NL. Clinical spectrum of alimentary tract duplication in children. Acta Paediatr Taiwan 2004;45(2):85-8.
6. Karnak I, Ocal T, Senocak ME, Tanyel FC, Büyükpamukçu N. Alimentary tract duplications in children: report of 26 years' experience. Turk J Pediatr 2000;42(2):118-25.
7. Raffensperger J. Duplications of alimentary tract. Swenson's Pediatric Surgery. 5th ed. Connecticut: Lange; 1990. p. 579-85.
8. Inoue Y, Nakamura H. Adenocarcinoma arising in colonic duplication cysts with calcification: CT findings of two cases. Abdom Imaging 1998;23(2):135-7.
9. Cavallaro G, Arena R, D'Ermo G, Basile U, Polistena A, Scorsi A, et al. Cystic duplication of transverse colon: an unusual case of abdominal pain and bowel obstruction. G Chir 2010;31(5):236-8.
10. Jancelewicz T, Simko J, Lee H. Obstructing ileal duplication cyst infected with Salmonella in a 2-year-old boy: a case report and review of the literature. J Pediatr Surg 2007;42(5): E19-21.
11. Martins JL, Cury EK, Petrilli AS, Martins EC, Neto G. Cecal duplication causing a disappearing abdominal mass in an infant. J Pediatr Surg 2001;36(10):1581-3.
12. Grosfeld JL, Boles ET Jr, Reiner C. Duplication of pylorus in the newborn: a rare cause of gastric outlet obstruction. J Pediatr Surg 1970;5(3):365-9.
13. Tang XB, Bai YZ, Wang WL. An intraluminal pyloric duplication cyst in an infant. J Pediatr Surg 2008;43(12):2305-7.
14. Holcomb GW 3rd, Gheissari A, O'Neill JA Jr, Shorter NA, Bishop HC. Surgical management of alimentary tract duplications. Ann Surg 1989;209(2):167-74.