Axial Torsion and Gangrene of Meckel's Diverticulum: The First Infantile Case Report

Meckel Divertikülünün Aksiyal Torsiyonu ve Gangreni: İlk Bebek Olgu Sunumu

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ABSTRACT Axial torsion of Meckel's diverticulum (MD) is very rare and has been reported in only 9 children and 23 adults within the past 40 years. Herein, we report the first known infant case of axial torsion and gangrene of MD. A 6-month-old male patient presented with acute abdomen. Ultrasonography showed a perihepatic cystic mass and minimal free fluid. Exploratory laparotomy revealed a gangrenous MD measuring 7 × 6 cm due to axial torsion. MD wedge resection was performed and the patient was asymptomatic during 2 years of follow-up. MD and its torsion and gangrene should be considered in surgical cases of acute abdomen.

Key Words: Meckel diverticulum; gangrene

ÖZET Meckel divertikülünün (MD) aksiyel torsiyonu ve gangreni oldukça nadirdir ve son 40 yılda çocuklarda sadece 9, yetişkinlerde de 23 olgu bildirilmiştir. Bu makalede, ilk bebek MD aksiyel torsiyonu ve gangreni olgusu sunulmuştur. Altı aylık erkek hasta, akut karın bulguları ile getirildi. Ultrasonografide perihepatik kistik kitle ve minimal serbest sıvı saptandı. Eksploratris laparatomide, 7 × 6 cm boyutlarında aksiyel torsiyon ve gangren gelişmiş MD saptandı ve "wedge" rezeksiyon ile çıkarıldı. Hastada 2 yıllık izlem boyunca sorun olmadı. MD torsiyonu ve gangreni akut karın olgularında akılda tutulmalıdır.

Anahtar Kelimeler: Meckel divertikülü; gangren

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Meckel's diverticulum (MD), originating as a result of non-closure of the Vitelline duct, is the most common (2%) congenital gastrointestinal system anomaly. It may be accompanied by esophageal atresia, anal atresia, and neurologic and cardiovascular malformations. Although most cases are silent, they may become symptomatic (3.7-6.4%) due to bleeding, obstruction, and inflammation.[1-5] In clinical practice, MD mostly presents with gastrointestinal bleeding (10.9-38.9%), diverticulitis (12.7-30.9%), and intestinal obstruction (26.6–53.4%).[1,2] MD gangrene due to axial torsion is the least commonly reported complication among children and adults; we had previously reported one case in a child. In the literature, 9 cases in children and 23 cases in adults have been reported over the last 40 years.[6-10] Here, we presented our second case of this rare complication in a 6-month-old boy, who was the first reported infant case.

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CASE REPORT

A 6-month-old male patient presented to our clinic with restlessness, abdominal distension, inability to defecate, bilious vomiting, and fever for the last 5 days. Physical examination revealed severe abdominal distension limiting deep palpation and findings of acute abdomen. Axillary body temperature was 40.1°C. An upright abdominal plain x-ray revealed air-fluid levels and dilated intestinal loops (Figure 1). Ultrasonography showed a cystic perihepatic mass and minimal free fluid. The leukocyte count was 48,300/mm^3. Explorative laparotomy revealed a gangrenous 7x6 cm MD originating from the ileal wall that was rotated 540 degrees counterclockwise around its own root and 20 cm from the ileocecal junction (Figure 2). MD wedge resection was performed. There were no postoperative complications and the patient was discharged on postoperative day 7. Histopathology confirmed a full-thickness necrotic degenerated MD. The patient was asymptomatic during 2 years of follow-up.

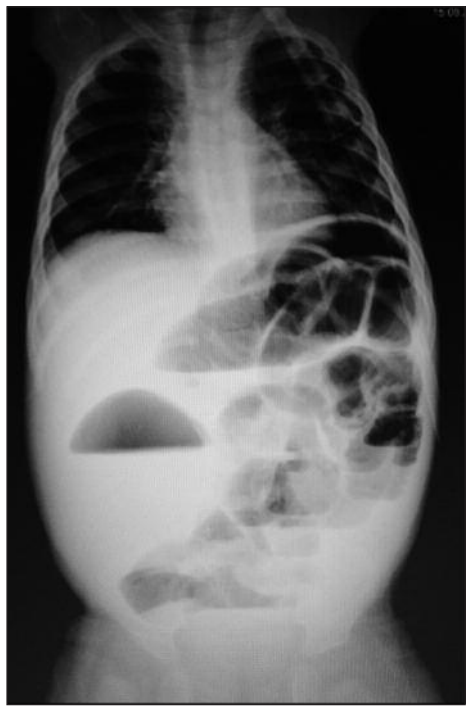

FIGURE 1: Air-fluid levels and dilated intestinal loops in the upright abdominal plain x-ray.

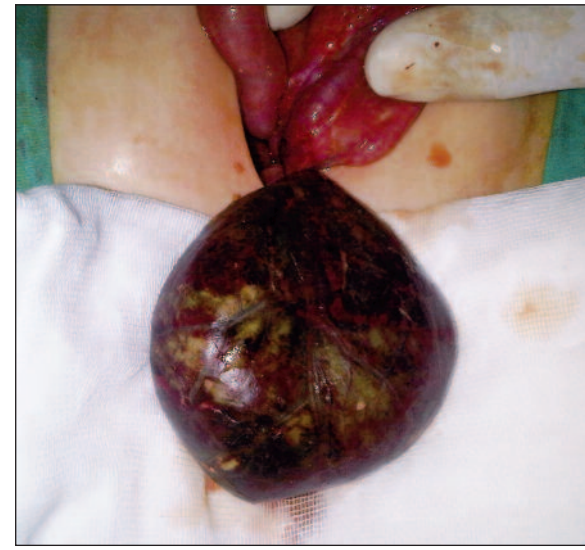

FIGURE 2: Gangrenous torsion of Meckel's diverticulum in an infant.
(See color figure at http://tipbilimleri.turkiyeklinikleri.com/)

The child's father gave consent to the use of medical photographs and recordings of his son's medical treatment for teaching, research purposes and publishing.

DISCUSSION

Complications of MD include intestinal strangulation, intussusception, incarcerated hernia, and neoplastic transformation. Gangrene of the MD due to axial torsion is a very rare complication[6-10] and may present with non-specific abdominal pain, nausea, and vomiting, particularly in children. It is quite difficult to diagnose MD preoperatively based on radiologic methods. MD may be visualized using a radioisotope study with Tc-99m pertechnetate with high sensitivity and specificity.[4,5] Computerized tomography and ultrasonography may detect complications in symptomatic cases.

Ten children (ages ranging from 6 months to 14 years of age; 2 girls, 8 boys), including our current case and our previously reported case, have presented with non-specific abdominal pain, anorexia, nausea, and vomiting and have undergone emergency surgery due to acute abdomen with initial diagnoses of a strangulated ileus, appendicitis, intestinal obstruction, duplication cyst, and perforated appendicitis with abscess formation.[6-10] Axial

torsion of MD could not be diagnosed preoperatively in all cases; however, in some cases, ultrasonography showed an incompressible cystic mass and computerized tomography revealed a cystic mass without contrast uptake, as well as a dilated intestine-like mass.[6-10] Therefore, computerized tomography should be done in the acute abdomen cases of suspected MD or its complication. A scintigraphic examination was not performed in all cases.[6-10] We feel that, particularly in gangrenous cases, it is hard to obtain results from scintigraphy, which is already difficult to perform on an emergency basis.

The youngest case reported up to date was a 6-year-old boy reported by Limas et al.[10] He was operated with the diagnosis of acute appendicitis with acute abdomen signs and symptoms (abdominal pain, nausea, fever, high white blood cell, abdominal tenderness) similar to our case. However, our 6-month-old boy case is the first infant case of MD with axial torsion and gangrene.

The treatment for symptomatic MD is surgery. However, preoperative diagnosis is very difficult to achieve, as the symptoms mimic many other disorders. The patients frequently undergo surgery with the initial diagnosis of acute abdomen and get their diagnosis.

MD and its complications should be considered in surgical cases of acute abdomen, particularly in cases where there is no specific pathology explaining the clinical condition. Although the postoperative outcome is usually good, preoperative diagnosis of MD is usually relatively indefinable, both at clinical and imaging examination. Delay in the diagnosis of a complicated MD can lead to significant morbidity and mortality. Therefore, a high index of suspicion is warranted to a correct and quick diagnosis of MD, valuable particularly in patients with atypical presentation. Torsion and gangrene of MD may surprisingly be present in cases with acute abdomen.

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