Acute abdomen due to Meckel's diverticulitis with synchronous inflammatory myofibroblastic tumor in the terminal ileum: A case report

Burak Dinçer, Sinan Ömeroğlu, Onur Güven, Mustafa Fevzi Celayir, Uygar Demir

Department of General Surgery, Şişli Hamidiye Etfal Training and Research Hospital, Istanbul-*Türkiye*

ABSTRACT

Meckel's diverticulum (MD) is the most common congenital anomaly of the gastrointestinal system, occurring in approximately 2% of the population. It is rare for MD to be symptomatic or complicated in adulthood. In this case report, we describe a patient who was admitted to the clinic with Meckel's diverticulitis, which had fistulized to the anterior abdominal wall, and was incidentally found to have an ileal inflammatory myofibroblastic tumor (IMT) on radiological imaging. A 46-year-old male patient presented to the emergency department with abdominal pain. Physical examination revealed localized guarding on the right side of the umbilicus. Blood tests showed elevated acute-phase reactants, including a white blood cell count of 13,800/μL, and C-reactive protein (CRP) level of 165 mg/L. Abdominal computed tomography demonstrated Meckel's diverticulitis fistulizing to the anterior abdominal wall and a polypoid structure in the ileum distal to the MD. The patient underwent emergency surgery, during which segmental ileal resection and ileocolic anastomosis were performed. On the fourth postoperative day, the patient developed an anastomotic leak. Re-laparotomy, right hemicolectomy with end ileostomy, and mucous fistula creation were subsequently performed. Pathological examination of the resected ileum from the initial surgery revealed a benign IMT distal to the MD. The patient was discharged on the 40th postoperative day after developing a surgical site infection following the second surgery. The end ileostomy was closed six months later. In this case, it appears that the ileal IMT located distal to the MD may have caused intermittent intestinal obstruction, fecal stasis, and the development of Meckel's diverticulitis. Furthermore, a detailed examination of the patient's history, laboratory results, and radiologic tests may contribute to the detection of incidental pathologies and influence treatment choices.

Keywords: Acute abdomen; enterocutaneous fistula; inflammatory myofibroblastic tumor; Meckel's diverticulitis.

INTRODUCTION

Meckel's diverticulum (MD) is the most common congenital gastrointestinal anomaly, with a prevalence ranging from 0.3% to 2.9% in the general population. MD typically presents with clinical symptoms more frequently in childhood, and only 4% of adults with MD develop symptoms.[1,2] In the adult population, complications include hemorrhage, bowel obstruction, diverticulitis, perforation, and malignancy.[1] Its clinical course often overlaps with other acute abdominal diseases. Although malignancy in Meckel's diverticulum is rare, both primary (neuroendocrine tumor, gastrointestinal stromal tumor, adenocarcinoma, and pancreatic epithelial neoplasia) and secondary malignancies (serous carcinoma originating from the uterus or ovary, and peritoneal mesothelioma) can occur.[1,3] Additionally, it has been noted that some patients with Meckel's malignancy also develop other primary malignancies at some point in their lives.[4] MD, whose clinical course is highly variable, can also be associated with benign tumoral lesions, such as ileal polyps in different locations.[2,5]

The World Health Organization classifies inflammatory myofibroblastic tumor (IMT) as an uncommon, intermediate mes-

Cite this article as: Dinçer B, Ömeroğlu S, Güven O, Celayir MF, Demir U. Acute abdomen due to Meckel's diverticulitis with synchronous inflammatory myofibroblastic tumor in the terminal ileum: A case report. Ulus Travma Acil Cerrahi Derg 2024;30:764-767.

Address for correspondence: Burak Dinçer

Department of General Surgery, Şişli Hamidiye Etfal Training and Research Hospital, Istanbul, Türkiye

E-mail: burak-dincer@hotmail.com

Ulus Travma Acil Cerrahi Derg 2024;30(10):764-767 DOI: 10.14744/tjtes.2024.82091

Submitted: 18.02.2024 Revised: 01.08.2024 Accepted: 06.08.2024 Published: 07.10.2024

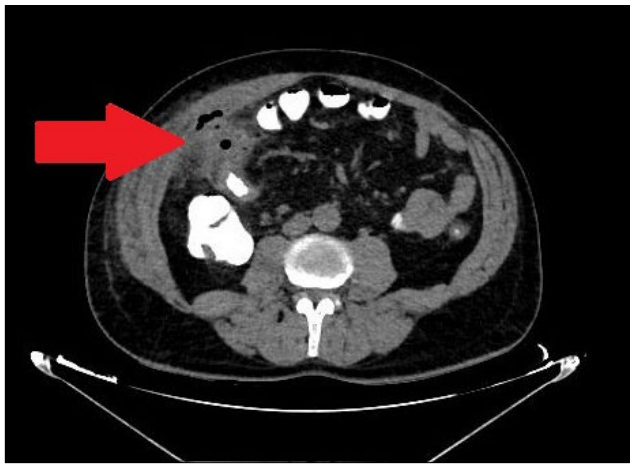

Figure 1. Meckel's diverticulitis fistulizing to the anterior abdominal wall, as seen on computed tomography (CT) scan (arrow).

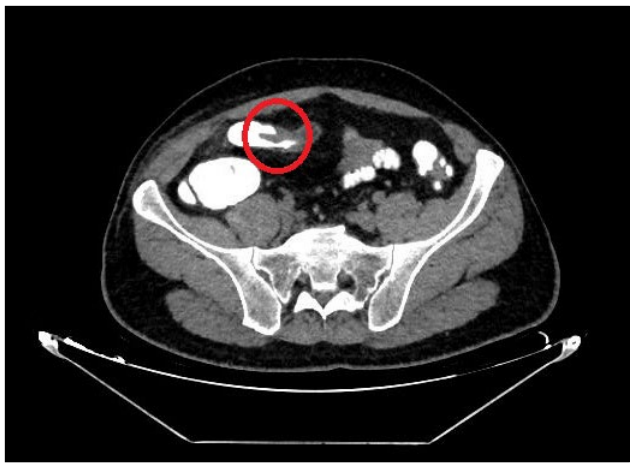

Figure 2. Pedicle of the polypoid lesion, as seen on CT (circled).

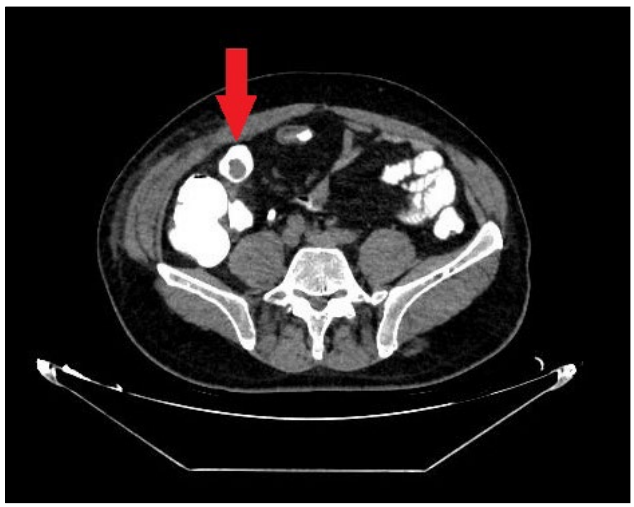

Figure 3. Ileal polypoid lesion, as seen on CT (arrow).

enchymal lesion.[6,7] IMT is generally seen in the pulmonary system, with the small bowel accounting for 1% of the affected sites.[6] Gastrointestinal IMT presents with symptoms such as anemia, weight loss, fecal occult blood positivity, abdominal pain, obstruction, or intussusception.[8] A limited number of cases of ileal IMT causing intussusception have been reported in the literature.[7]

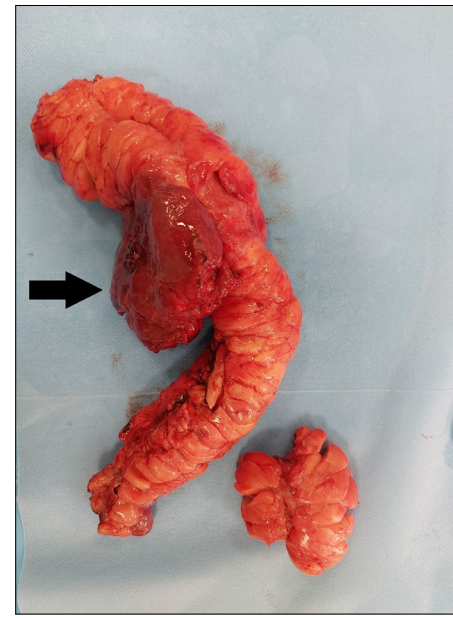

Figure 4. Resected ileal segment from the first surgery, with Meckel's diverticulitis indicated by the arrow.

We aim to present these two extremely rare conditions, which may occur synchronously and cause acute abdomen, and to emphasize the importance of preoperative imaging and thorough abdominal exploration during surgery.

CASE REPORT

A 46-year-old male patient with no prior comorbidities or surgical history presented with abdominal pain. He had a previous hospitalization with a diagnosis of Meckel's diverticulitis one month prior. Surgical intervention was recommended, but the patient declined surgery at that time. Vital signs were within normal limits. Physical examination revealed localized guarding in the right side of the umbilicus. Blood tests showed elevated acute-phase reactants (white blood cell count: 13,800/μL, C-reactive Protein: 165 mg/L). Contrast-enhanced abdominal computed tomography (CT) scan confirmed Meckel's diverticulitis in the distal ileum with a fistula to the anterior abdominal wall (Fig. 1). Incidentally, a polypoid lesion causing partial obstruction was identified distal to the MD (Figures 2 and 3). The patient was initiated on intravenous antibiotics and fluid therapy, followed by surgical intervention. During surgical exploration, a fistulized Meckel's diverticulitis with a connection to the anterior abdominal wall was located 40 cm proximal to the ileocecal valve (Fig. 4). The ileum distal to the MD, including the ileocecal valve, exhibited severe edema, inflammation, and dilation. A segmental ileal resection with an end-to-side ileocolic anastomosis to the ascending colon was performed.

On the fourth postoperative day, a suspicion of anastomotic leak prompted an oral contrast-enhanced CT scan. The CT scan revealed contrast extralumination from the ileocolic anastomosis. As the patient was not a candidate for non-operative management, a repeat laparotomy was performed. The ascending colon was severely inflamed and ischemic. Consequently, a right hemicolectomy, end ileostomy, and mucus fistula creation of the transverse colon were performed. The patient required long-term intravenous antibiotics due to postoperative wound and intra-abdominal infection. He was discharged on the 40th postoperative day. Six months later, the ileostomy was closed. Pathological examination revealed a 3x1.5 cm perforated Meckel's diverticulitis in the middle portion of the resected ileal segment. Additionally, a 6x1.5 cm benign inflammatory myofibroblastic tumor was identified distal to the MD.

DISCUSSION

MD is the most common anomaly of the gastrointestinal system and often presents symptomatically in childhood. Fifty percent of patients undergoing surgery for MD are younger than 10 years old, while complications due to MD in adulthood are very rare.[1,2] Literature on MD and its complications in adult patient groups generally consists of case reports and series, and the optimal approach to MD in adults is controversial. While surgical resection is favored in cases of Meckel's diverticulitis, there is insufficient evidence to recommend routine surgical resection for incidentally detected MDs during abdominal surgery.[9]

There is no definitive consensus on the factors that cause complications in patients with MD. Fecal stasis or secretions from ectopic mucosa are believed to play a role in the development of complications such as Meckel's diverticulitis. The simultaneous occurrence of conditions such as fecaliths, parasite infestation, and neoplasia, which obstruct the diverticulum orifice and lead to fecal stasis, in most reported cases of Meckel's diverticulitis supports this hypothesis.[10-12]

IMTs are borderline tumors that commonly occur in the lungs but are extremely rare in the gastrointestinal tract. Cases of intestinal obstruction caused by IMTs have been described in the literature.[6-8] Meckel's diverticulitis in adults is a rare condition and is often reported to be associated with other pathologies.[2,5,9,13] However, there are no documented cases of ileal IMT coexisting with Meckel's diverticulitis in the literature, making our report the first to describe this association.

In the present case, while Meckel's diverticulitis was the primary cause of symptoms, an IMT was discovered in the distal ileum. Careful examination of radiological images and standard surgical abdominal exploration in patients with acute abdomen are important for detecting not only the primary pathology but also potential additional pathologies.

CONCLUSION

Meckel's diverticulitis in adults is a very rare condition that can be associated with other pathologies. Ileal IMT, which can lead to intestinal obstruction, may coexist with Meckel's diverticulitis. A detailed preoperative evaluation that considers the possibility of co-occurring pathologies can influence the surgical plan.

Peer-review: Externally peer-reviewed.

Authorship Contributions: Concept: B.D., S.Ö.; Design: B.D., S.Ö., O.G.; Supervision: M.F.C., U.D.; Resource: B.D., S.Ö., O.G., M.F.C., U.D.; Materials: B.D., S.Ö., O.G., M.F.C., U.D.; Data collection and/or processing: B.D., S.Ö., O.G., M.F.C., U.D.; Analysis and/or interpretation: B.D., S.Ö., O.G., M.F.C., U.D.; Literature search: B.D., S.Ö., O.G.; Writing: B.D., S.Ö.; Critical reviews: B.D., S.Ö., O.G., M.F.C., U.D.

Conflict of Interest: None declared.

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

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OLGU SUNUMU - *ÖZ*

Meckel divertikülitine bağlı akut batın olgusunda terminal ileumda senkron inflamatuar myofibroblastik tümör: Olgu sunumu

Burak Dinçer, Sinan Ömeroğlu, Onur Güven, Mustafa Fevzi Celayir, Uygar Demir

Şişli Hamidiye Etfal Eğitim ve Araştırma Hastanesi, Genel Cerrahi Kliniği, İstanbul, Türkiye

Meckel divertikülü (MD) gastrointestinal sistemin en sık görülen konjenital anomalisi olup popülasyonda yaklaşık %2 oranında görülmektedir. Erişkin dönemde MD'nin semptomatik veya komplike olması nadirdir. Bu olgu sunumunda batın ön duvarına fistülize Meckel divertiküliti ile kliniğe başvuran ve radyolojik görüntülemelerinde insidental olarak ileal inflamatuar myofibroblastik tümör (IMT) saptanan bir hasta anlatılacaktır. Kırkaltı yaşında erkek hasta acile karın ağrısı şikayeti ile başvurdu. Fizik muayenede umblilikus sağ tarafında lokalize defans saptandı. Kan tetkiklerinde akut faz reaktanlarının arttığı görüldü (Beyaz küre: 13,800/μL, CRP: 165 mg/L). Batın bilgisayarlı tomografisinde batın ön duvarına fistülize Meckel divertiküliti ve MD'nin distalinde ileum içerisinde polipoid yapı olduğu izlendi. Hasta acil ameliyata alındı. Segmenter ileum rezeksiyonu ve ileokolik anastomoz yapıldı. Ameliyat sonrası 4. günde anastomoz kaçağı gelişen hastaya re-laparotomi, sağ hemikolektomi, uç ileostomi ve müköz fistül açılması ameliyatı yapıldı. İlk ameliyatta rezeke edilen ileum segmentinin patolojik incelemesinde MD'nin distalinde benign İMT olduğu görüldü. Yara yeri ve intraabdominal enfeksiyon nedeniyle uzamış intravenöz antibiyoterapi uygulanan hasta ameliyat sonrası 40. günde taburcu edildi. Altı ay sonra hastanın ileostomisi kapatıldı. Olgumuzda ileum yerleşimli İMT'nin aralıklı intestinal obstrüksiyon ataklarına, fekal staza ve buna bağlı olarak Meckel divertiküliti gelişimine neden olduğu düşünülebilir. Ek olarak hastanın hikayesinin, laboratuvar ve radyolojik testlerinin dikkatli incelenmesinin tedavi üzerine etkili olabilecek insidental patolojilerin saptanmasında katkısı olacağı söylenebilir.

Anahtar sözcükler: Akut batın; enterokütanöz fistül; inflamatuar myofibroblastik tümör; meckel divertiküliti.

Ulus Travma Acil Cerrahi Derg 2024;30(10):764-767 DOI: 10.14744/tjtes.2024.82091