Case Report / Olgu Sunumu

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A Case with Multiple Systemic Inflammatory Syndrome Presenting with Acute Appendicitis Symptoms

Akut Apandisit Semptomları ile Başvuran Çoklu Sistemik Enflamatuvar Sendromlu Bir Olgu

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Abstract

Coronavirus disease-2019-associated pediatric multisystem inflammatory disease has been defined as a severe disease that causes fever, abdominal pain, hypotension, and myocardial dysfunction in children with severe acute respiratory syndrome-coronavirus-2 infection. However, some multiple systemic inflammatory syndrome (MIS-C) cases progress to multi-organ failure requiring intensive care follow-up. In the patient who had severe abdominal pain, vomiting and high fever and was diagnosed with acute appendicitis in the emergency room, the diagnosis of MIS-C was considered during the follow-up, and parasitic infestation, which is one of the rare etiological causes of acute appendicitis, was detected.

Keywords: MIS-C, COVID-19, acute appendicitis, pediatric patient

Öz

Koronavirüs hastalığı-2019 ilişkili çocuk multisistem enflamatuvar hastalık, şiddetli akut solunum sendromu-koronavirüs-2 enfeksiyonu geçiren çocuklarda ateş, karın ağrısı, hipotansiyon ve miyokardiyal işlev bozukluğuna yol açan şiddetli bir hastalık olarak tanımlanmıştır. Bununla birlikte bazı MIS-C olguları, yoğun bakım takibi gerektirecek çoğul organ yetmezliğine ilerlemektedirler. Şiddetli karın ağrısı, kusma ve yüksek ateş şikayeti olan, acil serviste akut apandisit tanısı konulan olguda izlemde MIS-C tanısı düşünülmüş ve akut apandisitin ender etiyolojik nedenlerinden paraziter enfestasyonu tespit edilmiştir. Bu olgu çoğul sistemik enflamatuvar sendromun (MIS-C) gastrointestinal sistem tutulumunun nadir bir bulgusu olan akut apandisit ile tanı alması ve akut apandisitin ender etiyolojik nedenlerinde paraziter enfestasyonu tespit edilmesi nedeniyle sunulmuştur.

Anahtar Kelimeler: MIS-C, COVID-19, akut apandisit, çocuk hasta

Introduction

In December 2019, an epidemic of pneumonia of unknown cause occurred in Wuhan, China's Hubei Province, and it was understood that a new type of coronavirus caused the disease. The new virus was named severe acute respiratory failure syndrome-coronavirus-2 (SARS-CoV-2), and the disease it caused was named Coronavirus disease-2019 (COVID-19). The disease spread all over the world in a short time, causing a pandemic.[1] COVID-19, a severe disease that causes fever, abdominal pain, hypotension, and myocardial dysfunction in children infected with SARS-CoV-2, was described in Europe in April 2020. Multiple organ failure and the need for intensive care have been observed in some cases.[2,3] While this syndrome was defined as the pediatric inflammatory multisystem syndrome (PIMS-TS) associated with transient SARS-CoV-2 in Europe, it was named as COVID-19-associated multiple systemic inflammatory syndrome (MIS-C) by the Center for Disease Control and Prevention (CDC).[4,5]

Appendicitis is one of the most common causes of abdominal pain and emergency gastrointestinal surgery. Fecal stasis, fecalitis and lymphoid hyperplasia are frequently involved in the etiology of appendicitis. Intestinal parasites and tumors are rarely found in the etiology of appendicitis.[6] If the emergency

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surgical intervention of acute appendicitis is delayed, the clinic of simple appendicitis may result in perforation and the delay may increase morbidity and mortality.[7]

This case is presented to emphasize both the differential diagnosis of acute appendicitis with refractory fever and MIS-C with gastrointestinal findings, and the fact that *Enterobius vermicularis* is a very rare etiologic cause of acute appendicitis.

Case Report

An 8.5-year-old girl, whose personal and family history was unremarkable, was admitted to the emergency department with the complaints of severe abdominal pain, vomiting and high fever. In the physical examination, her abdomen was tender and the rebound finding was positive. In the computed tomography imaging, the appendix was measured as 7.3 mm at its thickest point. The pathology of the appendectomy material of the patient who was operated with a preliminary diagnosis of acute appendicitis was reported as follows: "It is chronic inflamed appendix tissue with parasite fragments in the lumen, and morphological findings suggest *Enterobius vermicularis*". Despite the antibiotic treatment, her fever continued, and the patient, who started to have respiratory distress and progressed on the 3rd postoperative day, was admitted to the intensive care unit.

In the intensive care examination, she had toxic-looking, she was conscious, her respiratory was tachypneic (56/min) and dyspneic, pulmonary breath sounds were normal, she had tachycardia (154/min), weak heart rate, arterial blood pressure of 96/56 mmHg, and gallo rhythm. Her abdomen was tender, the liver was palpated as 3 cm, and maculopapular rash in the lower and upper extremities, non-purulent conjunctivitis, and strawberry tongue were detected. In echocardiography, mild mitral insufficiency and ejection fraction of 58% were detected. Laboratory results were as follows: SARS-CoV-2 polymerase chain reaction: Negative, COVID-19 (SARS-CoV-2) IgG-IgM: positive, leukocyte: 6.400 (/µL), hemoglobin: 11.4 (g/dL), platelet: 149 (thousand/µL), lymphocyte: 700 (/µL), activated partial thromboplastin time: 26.3 (sec), pentylenetetrazol: 14.7 (sec), international normalised ratio (INR): 1.25 (sec), fibrinogen: 435.9 (mg/dL), alanine transaminase: 71 (U/L), aspartate transaminase: 42 (U)/L, total protein: 5.7 (g/dL), albumin: 2.4 (g/dL), sodium: 133 (mmol/L), potassium: 2.8 (mmol/L), ferritin: 1.922 (mg/L), troponin: I 0.210 (mg/L), brain natriuretic peptide: 533 (pg/ML), D-dimer: 8.4 (ng/mL), sedimentation: 59 (mm/h), C-reactive protein (CRP): 128 (mg/L), procalcitonin: 10.74.

The patient was started on oxygen with a high-flow nasal cannula. Oral feeding was discontinued, and limited intravascular fluid was planned due to the loading findings. MIS-C was considered due to clinical findings, test results, non-reducing fever and multi-organ failure. When arterial blood pressure continued to decrease (90/50 mmHg) to the hypotensive limit for age, the mean arterial blood pressure was measured as 63 mmHg, and circulatory disorder developed, adrenaline infusion was started. The vasoactive inotrope score was calculated as 5. Antibiotherapy was revised, Intravenous immunoglobulin (IVIG), corticosteroid and enoxaparin sodium treatment were administered. On the second day of her hospitalization, her fever decreased, her respiratory distress regressed, and her circulatory disorder improved. She was transferred to the ward without complications. "Patient consent information" was obtained from the legal representative of the patient.

Discussion

The pathogenesis of multisystemic inflammatory syndrome is still unknown. The fact that these cases usually occur some time after SARS-CoV-2 infection suggests that the cause of the disease may not be the direct effect of the virus, but the reason has not been fully elucidated.[8] Our case was followed up in the intensive care unit and diagnosed with MIS-C because she had high fever lasting more than 5 days, microorganism could not be grown in cultures, she had SARS-CoV-2 serology positivity, she had high laboratory test values of lymphopenia, hypoalbuminemia, hyponatremia, hyperfibrinogenemia, aspartate aminotransferase, alanine aminotransferase, lactate dehydrogenase, INR, pentylenetetrazole, D-dimer, ferritin, CRP, sedimentation, procalcitonin, pro-BNP, troponin-I and multiple organ failure. Our case met both the Ministry of Health MIS-C case definition[9] and CDC's MIS-C diagnostic criteria.[4]

The clinical symptoms of MIS-C manifest themselves in a wide spectrum affecting many systems. Most affected children are previously healthy and have no history of underlying disease.[10] Similar to the studies in the literature, our patient had no history of underlying disease and underwent acute appendectomy 3 days ago. The etiology of appendicitis often includes fecal stasis, fecalitis, and lymphoid hyperplasia. Intestinal parasites, tumors, radiological studies using barium, undigested vegetable scraps and fruit seeds are also rarely found in the etiology of appendicitis.[6] Altun et al.[11] found that 1.8% of 660 acute appendectomy materials had parasite infestation in their histopathological diagnosis, and 75% of them were *Enterobius vermicularis*. In the literature, as in the study of Altun et al.[11] it was important to detect *Enterobius vermicularis* in our patient as a very rare etiological cause of acute appendicitis.

MIS-C is a systemic disease involving multiple systems, and the treatment and follow-up of affected children requires multidisciplinary coordination. The American Society of

Rheumatology MIS-C treatment recommendation should be applied according to the clinical condition of the patient. Antibiotic, IVIG, and antithrombotic therapies are appropriate for patients with moderate to severe symptoms.[12] Our patient was accepted as severe MIS-C with clinical findings and laboratory results. A 15-year-old female patient with similar complaints, as our case, was followed up by Aslan et al.[13]

MIS-C was considered because her fever continued after appendectomy operation. Unlike our case, anakinra and plasmapheresis treatment was applied because it did not respond to IVIG and steroid treatment. According to the MIS-C treatment guidelines, appropriate antibiotic, inotropic drug infusion, IVIG, glucocorticoid and antithrombotic treatment was administered to our patient.

In multisystemic inflammatory syndrome, clinical findings manifest themselves in a wide spectrum including many systems. In a multicenter study by Feldstein et al.[14], gastrointestinal system complaints were found in 92% of 186 MIS-C cases, and acute appendicitis was found in only two (1%) cases. In a similar study conducted by Yilmaz Ciftdogan et al.[15] in a multicenter study with 101 MIS-C patients, gastrointestinal symptoms were found in 80.2%, but acute appendicitis could not be detected.

Similar to the studies of both Feldstein et al.[14] and Yilmaz Ciftdogan et al.[15] in the literature, either no acute appendicitis was detected in MIS-C patients or it was detected very rarely. It is noteworthy that our case was diagnosed with acute appendicitis, which is a rare finding of gastrointestinal system involvement of MIS-C, and that parasitic infestation was detected in rare etiological causes of acute appendicitis. Differential diagnosis should be made with acute appendicitis with treatment-resistant fever and MIS-C with gastrointestinal findings.

Ethics

Informed Consent: "Patient consent information" was obtained from the legal representative of the patient.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Concept: A.K., S.K., Design: A.K., S.K., Data Collection or Processing: A.K., S.K., Analysis or Interpretation: A.K., S.K., Literature Search: A.K., S.K., Writing: A.K., S.K.

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