HEME : Health and Medical Journal
pISSN : 2685 – 2772
eISSN : 2685 – 404x
Available Online at : https://jurnal.unbrah.ac.id/index.php/heme/issue/view/61

A Case Report: Unusual Manifestation of MIS-C with Acute Abdomen Caused by Gut Perforation

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Abstract

Background: Gastrointestinal symptoms are common presenting features of multisystem inflammatory syndrome in children (MIS-C) and can overlap with other abdominal symptoms. **Case Report**: We reported A case of a 2-year-old girl who was hospitalized with MIS-C and had abdominal pain symptoms which got worse day by day. Abdominal x-ray revealed gut perforation and she was consulted by the pediatric surgeon and then underwent an emergency exploratory laparotomy and intraoperative findings showed a perforation in the caecum area. **Conclusion**: MIS-C can induce acute gut perforation and in the pandemic era MIS-C should be considered in the differential for a patient with acute abdominal symptoms.

Keywords : MISC, gut perforation, covid-19

Abstrak

Latar Belakang*: Gejala gastrointestinal merupakan gejala umum sindrom inflamasi multisistem pada anak-anak (MIS-C) dan dapat tumpang tindih dengan gejala akut abdomen lainnya.* ***Laporan Kasus****: Kami melaporkan kasus seorang anak perempuan berusia 2 tahun dengan diagnosis MIS-C dan memiliki gejala nyeri perut yang semakin memberat dari hari ke hari. Rontgen abdomen menunjukkan gambaran perforasi usus dan dikonsultasikan ke dokter bedah anak dan kemudian dilakukan laparotomi eksplorasi dengan temuan intraoperatif menunjukkan perforasi di area sekum.* ***Kesimpulan****: MIS-C dapat menyebabkan perforasi usus dan di era pandemi MIS-C harus dipertimbangkan sebagai diagnosis banding akut abdomen.*

Kata Kunci : *MISC, perforasi usus, infeksi covid-19*

I. INTRODUCTION

COVID-19 infection in children was initially considered not to cause severe clinical symptoms and rarely causes death. However, recent data show that children experience severe symptoms following COVID-19 infection due to an exaggerated inflammatory response known as multisystem inflammatory syndrome in children (MIS-C), which can lead to organ damage, causing disability, and even death. MIS-C has a diverse clinical spectrum due to hyperinflammatory processes in various organs and tissues, including the gastrointestinal system.[1,2]

Gastrointestinal manifestations were reported among 92% of MIS-C cases. Common clinical manifestations include diarrhea, abdominal pain and constipation, pancreatitis, hepatitis or hepatomegaly, gallbladder hydrops or edema, and other complications including ileitis, colitis, or mesenteric adenitis.[3] Gastrointestinal symptoms in MIS-C also often cause acute abdomen manifestation, and some even require surgical exploration.[4] Several case reports of MIS-C with acute abdomen generally have clinical appendicitis-like symptoms with intraoperative findings of a normal appendix or edematous appendicitis.[4,5,6] MIS-C cases with gut perforation are still rarely reported. To our knowledge, this is the first report of the initial presentation of MIS-C with severe inflammatory acute abdomen with diffuse peritonitis caused by gut perforation.

II. CASE REPORT

A 2-year-old previously healthy girl was referred from the district hospital to our hospital because of a high and prolonged fever with pancytopenia. The patient had a history of high fever 3 weeks before treatment, with a peak of 40 ^{o}C. The temperature only decreased shortly after antipyretic administration. The patient also had abdominal bloating 3 days before

hospitalization, accompanied by abdominal pain. The patient looked pale and lost her appetite. There was no history of bleeding, or diarrhea, and the micturition was within normal limits. Previously, the patient was hospitalized 1.5 months ago due to fever and red rash spreading over the trunk and extremities for 4 days before being discharged. In this current episode, the patient was readmitted 3 weeks ago due to prolonged fever and pancytopenia.

Based on the physical examination when admitted, the patient was severely ill, had tachycardia (145 x/minute), tachypnea (39 x/minute), had a high fever (39.5 ^{o}C), and blood pressure was 100/60 mmHg. The oral cavity was reddish with cracked lips. Reddish skin on palms and soles also with desquamation. The conjunctiva was pale. There was multiple lymph node enlargement palpable in the region of colli bilateral. The abdomen was flat and supple, with tenderness in the right hypochondrium area, no rebound tenderness, and bowel sound was normal. There is no liver and spleen enlargement.

FIGURE 1. THE PATIENT PICTURE; REDDISH ORAL CAVITY WITH CRACKED LIPS, PALMAR ERYTHEMA, AND SKIN DESQUAMATION

Data from the laboratory showed that the patient had pancytopenia. The patient tested negative for the SARS-CoV-2 RT-PCR, but the SARS-CoV-2 titer antibody was increased (>250 U/mL) and accompanied by an increase of inflammatory markers, as shown in Table 1. The patient has met the WHO criteria for MIS-C. She was treated with methylprednisolone 2 mg/kgBW/day, intravenous immunoglobulin (IVIG) 2 gr/kgBW, and a PRC transfusion was also given. Aspirin and enoxaparin admission were delayed due to thrombocytopenia (less than 50,000/ mm^3). The condition improved, and the fever declined rapidly, but the patient still complained of abdominal pain. Platelet count improved without transfusion, and low-dose aspirin, 3 mg/kgBW, along with prophylactic dose enoxaparin was initiated.

TABLE 1. THE LABORATORY FINDINGS OF THE PATIENT

		1st day of hospitalization	4th day of hospitalization	1 day before discharge
1	Haemoglobin	8 g/dL	10.2 g/dL	12.5 g/dL
2	Leukocyte	3,210/ mm^3	6,350/ mm^3	10,430/ mm^3
3	Thrombocyte	29,000/ mm^3	76,000/ mm^3	454,000/ mm^3
4	PT (Normal: 9.2-12.4)	11.9 seconds	11.7 seconds	12.1 seconds
5	aPTT (Normal: 21.1-28.5)	28.9 seconds	26.4 seconds	22.6 seconds
6	D-dimer	>10,000 ng/mL	5175 ng/mL	261 ng/mL
7	Troponin I	73 ng/L	14 ng/L	10 ng/L
8	Blood Sedimentation Rate (ESR)	60 mm	43 mm	13 mm
9	C-Reactive Protein (CRP)	87 mg/L	50 mg/L	27 mg/L
10	Procalcitonin	0.87 ng/mL	62.55 ng/mL	0.05 ng/mL
11	Ferritin	>1,200 ng/mL	>1,200 ng/mL	388 ng/mL
12	Albumin	3.3 g/dL	3.2 g/dL	3.4 g/dL
13	Ureum	11 mg/dL	15 mg/dL	12 mg/dL
14	Creatinin	0.3 mg/dL	0.2 mg/dL	0.32 mg/dL
15	SARS-CoV-2 RT-PCR	Negative		
16	Anti-SARS-CoV-2	>250 U/mL		

On the fourth day of hospitalization, the patient's condition was getting worse. The patient complained of diffuse abdominal pain, and vomiting, and looked very ill. On the abdominal examination, we found signs of acute abdomen, distended, hyper tympanic abdomen with a global reduction of bowel sounds, rebound tenderness, and signs of peritonitis. The patient had severe sinus tachycardia (190-210 x/minute) without fever. The patient developed hemodynamic failure and signs of shock (prolonged CRT, cold extremities, and hypotension). The shock was resolved after the patient had received about 20 mL/kgBW of ringer lactate and epinephrine drip, 0.1 mcg/kgBW/minute. The abdominal x-ray showed an image of the air-fluid levels and decreased air distribution in the lower abdomen, compatible with bowel obstruction characteristics. The patient was consulted by the pediatric surgeon and then underwent an emergency exploratory laparotomy. Aspirin and enoxaparin were temporarily stopped before surgery.

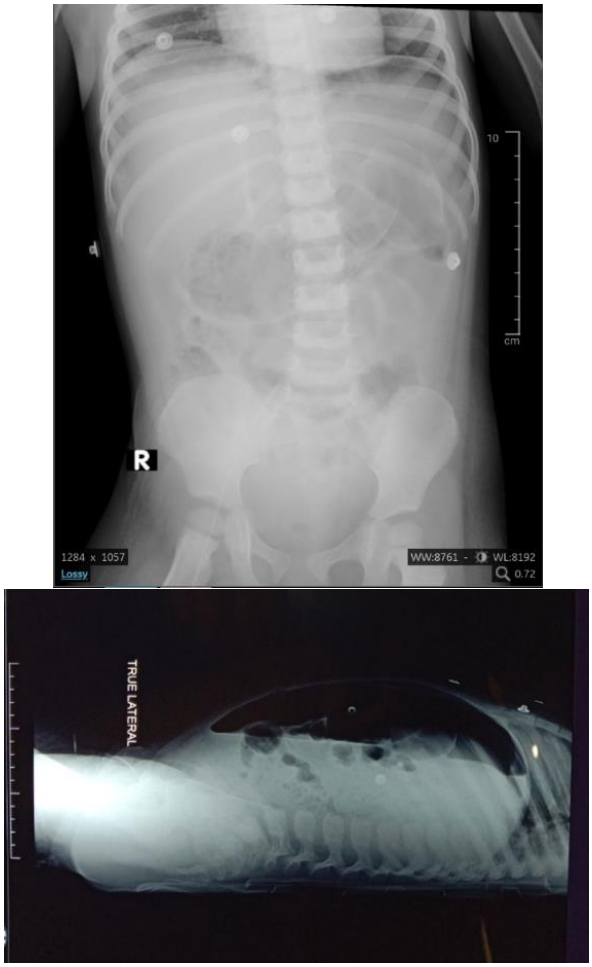

FIGURE 2. THE ABDOMINAL X-RAY SHOWED AN IMAGE OF THE BOWEL DISTENSION AND AIR-FLUID LEVELS

Intraoperative findings showed a perforation in the caecum area with a size of 0.5 cm x 0.5 cm, suture was performed on the perforated part, then the pediatric surgeon decided to do an ileostomy. The patient's clinical and hemodynamic condition improved after surgery, as well as the level of inflammatory markers. The patient was discharged after 18 days of hospitalization and was planned to close the ileostomy after 2 months.

FIGURE 3. THE INTRAOPERATIVE IMAGES SHOWED PERFORATION ON THE CAECUM

III. DISCUSSION

We present the case of an inflammatory acute abdomen with gut perforation associated with MIS-C. Even though abdominal symptoms are common manifestations of MIS-C, they rarely cause intestinal perforation. MIS-C patients with gastrointestinal symptoms often present with acute abdominal symptoms, and some cases require surgical exploration. This condition poses a new differential diagnosis for abdominal surgery in pediatric patients.[4]

Vecchio et al. reported that pancreatitis and abdominal fluid accumulation were more frequently observed in patients with MIS-C and indicated by pain, fever, and vomiting. Most of the 57 children with MIS-C (32 [56.1%]) had severe gastrointestinal

involvement, and conversely, MIS-C accounted for about half of the children with severe gastrointestinal manifestations and for all 6 cases of pancreatitis. In univariate analysis, MIS-C was associated with a higher risk of appendicitis (OR 4.71; 95% CI, 2.07-10.7), abdominal fluid accumulation (OR 22.9; 95% CI, 9.0-58.1), adenomesenteritis (OR 24.3; 95% CI, 11.8-49.7), and pancreatitis (OR 60.3; 95% CI, 6.9-525.7).[7]

Intraoperative findings of MIS-C-associated acute abdomen in several case reports revealed inflammatory appendicitis with abdominal fluid accumulation. However, there is also a missed diagnosis with the surgical finding of a normal appendix in some cases.[8]

Reports of MIS-C cases presenting as gut perforation are still limited. There are only a few case reports available, one of them was reported by Heza et al. reporting the occurrence of pneumatosis and gut perforation in a 17-year-old African-American girl morbidly obese. In this case, the patient presented with fever, abdominal pain, and shortness of breath. She progressed to hemodynamic failure and small bowel perforation approximately 1 week after admission.[9]

In another case, a 6-year-old girl with a positive history of COVID-19 5 weeks earlier, was admitted to a tertiary hospital with fever (up to 39 ^{o}C) and intermittent diarrhea that persisted for two days, as well as a 6-hour history of abdominal pain that started after single vomiting. A surgeon examined the child, identifying the classic clinical presentation of sudden appendicitis 3 hours before hospitalization followed by pelvic peritonitis, which appeared 2 hours later. Laparoscopy showed no inflammatory changes in the appendix but pronounced peritonitis with purulent (yellow, cloudy, and viscous) fluid of about 150 mL in the lower abdominal cavity.[10]

The mechanism of gut perforation in MIS-C is not fully understood. However, there are several theories linking the incidence of gut perforation in MIS-C to several factors (1) hypercoagulability in MIS-C, which leads to gut ischemia making it prone to perforation, and (2) gastrointestinal involvement in MIS-C is related to the presence of angiotensin-converting enzyme receptors in the gut and vascular epithelium, especially in glandular cells, absorptive enterocytes of the distal ileum and colon, which ultimately favor vasoconstriction, edema, glandular hyperplasia and a procoagulant state that leads to direct tissue damage.[4,11]

Lessons learned from the information obtained from this case were to be aware of abdominal symptoms, especially abdominal pain in MIS-C since there are possibilities of acute abdominal development that cause gut perforation.

IV. CONCLUSION

We should be aware of abdominal symptoms, especially abdominal pain in MIS-C since there are possibilities of acute abdominal development that cause gut perforation.

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