

Mucocutaneous and gastrointestinal involvement of MIS-A in a 67-year-old man: a case report

Sirs,

During the pandemic period, a multi-system inflammatory syndrome associated with SARS-CoV-2 was identified, firstly in children (MIS-C), and then in adults (MIS-A) (1). MIS-A is a COVID-19-related syndrome, generally presenting with a wide range of involvements (more frequently cardiovascular, gastrointestinal, or mucocutaneous) due to an imbalance in the immune system's response (2, 3).

We report a case of MIS-A, describing presentation, diagnosis, and management, highlighting the importance of thinking about MIS-A. As diagnostic and classification criteria and treatment guidelines for this new disease are not yet completely defined, it is important to analyse particularly the individual cases that can be found in clinical practice. Only in this way can we improve the patient's prognosis; indeed, early recognition and prompt therapy may result in a positive outcome (4).

In January 2023, a 67-year-old man (BMI 29.54), presented at the emergency department because of an episode of bloody diarrhoea and petechial-like skin lesions that were initially confined to the bilateral inner thighs and then extended also to the upper limbs (Fig. 1), without chest pain or shortness of breath. At the admission, he was anorectic, with an oxygen saturation of 97% (FiO₂ 21%) and a low blood pressure (103/67 mmHg) if compared with usual daily values. The patient had a medical history of hypertension and bilateral open-angle glaucoma; he had also been vaccinated for SARS-CoV-2 with 3 doses.

His recent history of illness began about a month earlier, with episodes of serotinous fever associated with mild weakness, muscle pain, ocular discomfort with lacrimation, decreased appetite, and weight loss, which spontaneously resolved within about 10 days. At the onset of the disease, chest x-ray was normal, and urine culture was negative for common germs and mycetes.

The petechial-like skin lesions appeared about two weeks after the onset of the fever. Blood tests performed in the Emergency Department revealed white blood cells 10190/mcL, haemoglobin 13.1 g/dL, CRP 8.45 mg/dL, lactate dehydrogenase 234 U/L, D-dimer 20961 ng/mL, fibrinogen 627 mg/dL, ferritin 709 mcg/L, ESR 49 mm/h and positive SARS-CoV-2 RT-PCR. On the same day, abdominal ultrasound and then also CT showed the presence of parietal thickening of the last four ileal arches with hyperaemic appearance of the mucosa, oedema, and hypervascularised appearance of the submucosa; mesenteric thickening, with vascular ectasia of small vascular branches, was also present.

According to the U.S. Center for Disease Control criteria, the clinical suspicion was MIS-A. The patient was thus admitted to the Department of Internal Medicine and then



Fig. 1. Petechial-like lesions of the lower limbs.

transferred to our Unit. On day 3, a rapid SARS-CoV-2 antigen test was negative.

Subsequently, immunomodulatory therapy with pulsed steroids (500 mg of 6 methylprednisolone for three consecutive days, then tapered) and intravenous Ig [IVIg] (400 mg/kg/day for 5 days) was initiated. Piperacillin/tazobactam and caspofungin were also administered to prevent superinfection. Parenteral nutrition was provided via a peripherally inserted central catheter. Echocardiography revealed only aortic sclerectasia and hypertensive heart disease. After 5 days, rectorrhagia ceased and angio-CT of the abdominal aorta documented general improvement (decreased vascular engorgement and wall thickness of the last intestinal arches). Antibiotic therapy was then stopped, and the patient was gradually shifted to oral feeding. Skin biopsy of a lesion in the left peritrochanteric region showed leukocytoclastic vasculitis (mild-moderate lymphocytes and neutrophil granulocytes infiltrate around vascular structures, numerous nuclear fragments in karyorexis and focal wall fibrinoid necrosis). In conclusion, even if studies on large cohorts, standardised by age, comorbidities, or clinical manifestations, are still necessary to define the best therapeutic strategy, the available data and this case argue in favour of using steroids in combination with IVIg (2 g/kg single/divided dose) and possibly biologicals (5-7).

F. PISTONE¹, MD
D. TESTA¹, MD
I. MARTELLI¹, MD
G. SCIASCIA¹, MD
G.M.L. RIZZELLI¹, MD
A. TRIPOLI², MD
S. DE MARCO², MD
P. MIGLIORINI¹, MD
A.G. TAVONI¹, MD

ORCID iD:

F. Pistone: 0009-0002-1273-5891
D. Testa: 0000-0002-5332-3781
I. Martelli: 0009-0006-3770-0301
P. Migliorini: 0000-0001-6433-4964

¹Clinical Immunology Unit,

²U.O. Medicina V, Azienda Ospedaliero-universitaria Pisana, Pisa, Italy.

Please address correspondence to:
Francesca Pistone

U.O. Immunoallergologia Clinica,
Università di Pisa, via Roma 67, 56126 Pisa, Italy.
E-mail: f.pistone2@studenti.unipi.it

Competing interests: none declared.

© Copyright CLINICAL AND

EXPERIMENTAL RHEUMATOLOGY 2024.

References

1. VOGEL TP, TOP KA, KARATZIOS C *et al.*: Multi-system inflammatory syndrome in children and adults (MIS-C/A): Case definition & guidelines for data collection, analysis, and presentation of immunization safety data. *Vaccine* 2021; 39(22): 3037-49. <https://doi.org/10.1016/j.vaccine.2021.01.054>
2. PARUMS DV: Editorial: Multisystem inflammatory syndrome in adults (MIS-A) and the spectrum of COVID-19. *Med Sci Monit* 2021; 27: e935005. <https://doi.org/10.12659/msm.935005>
3. PATEL P, DECUIR J, ABRAMS J, CAMPBELL AP, GODFRED-CATO S, BELAY ED: Clinical characteristics of multisystem inflammatory syndrome in adults: a systematic review. *JAMA Netw Open* 2021; 4(9): e2126456. <https://doi.org/10.1001/jamanetworkopen.2021.26456>
4. DIAKITE S, BOUSDIRA N, TACHON G, ACKERMANN F, GROH M, ROHMER J: Regression of coronary aneurysms with intravenous immunoglobulins and steroids for COVID-19 adult multisystem inflammatory syndrome. *JACC Case Rep* 2021; 3(4): 581-5. <https://doi.org/10.1016/j.jaccas.2021.01.012>
5. HOOKHAM L, FISHER C, MANSON JJ *et al.*: Understanding the diagnosis and management of multisystem inflammatory syndrome in adults (MIS-A) in the UK: results of a national Delphi process. *Clin Med (Lond)* 2022; 22(3): 266-70. <https://doi.org/10.7861/clinmed.2021-0700>
6. KUNAL S, ISH P, SAKTHIVEL P, MALHOTRA N, GUPTA K: The emerging threat of multisystem inflammatory syndrome in adults (MIS-A) in COVID-19: A systematic review. *Heart Lung J Crit Care* 2022; 54: 7-18. <https://doi.org/10.1016/j.hrtlng.2022.03.007>
7. CAITANE P, VOLPE A, CARDELLINO CS *et al.*: Multisystem inflammatory syndrome in an adult (MIS-A) successfully treated with anakinra and glucocorticoids. *Microorganisms* 2021; 9(7): 1393. <https://doi.org/10.3390/microorganisms9071393>