

CASE REPORT

COVID-19 associated Kawasaki-like multisystem inflammatory syndrome in an adult female with oral, ophthalmic and cardiac manifestation, a case report

Talayeh Mirkarimi^{1*}, Mohammad Salek²

1 Emergency Medicine department; Imam Ali Hospital, Alborz University of Medical Science, Taleghani Blvd, Karaj, Iran.

2 Oncology and Stem cell Transplantation Research Center, Shariati Hospital, Tehran University of Medical Sciences, Tehran, Iran.

Received: April 2023; Accepted: May 2023; Published online: 8 June 2023

Abstract: Kawasaki disease is an acute febrile disease with the vasculitis of small and medium vessels that mainly affects children under the age of five, usually occurring after a viral respiratory diseases. During the coronavirus disease-19 (COVID-19) pandemic, a significant increase was reported in the number of children with symptoms similar to Kawasaki disease, which is known as a multi-system inflammatory syndrome in children (MIS-C). A Kawasaki-like disease is rare in adults, especially in the context of COVID-19. We hereby present the case of A 41 year old female patient presented with complaints of bleeding from the gums and red and watery eyes, high fever, sore throat, weakness, and lethargy. She mentioned a recent contact with a COVID-19 patient. Covid-19 was confirmed by a positive reverse transcription polymerase chain reaction (RT-PCR) test. Other lab tests and clinical manifestations revealed inflammatory phenomena that fully complied with multi-system inflammatory syndrome in adults (MIS-A) criteria. The patient was treated with a possible diagnosis of MIS-A in the context of COVID-19 and was discharged in good general condition. COVID-19 rarely presents in adults without clear respiratory symptoms and in the form of multisystem inflammatory syndrome (MIS-A). Due to the possibility of irreversible complications, MIS-A requires special attention and early diagnosis and treatment.

Keywords: multisystem inflammatory syndrome, MIS-A, adult, COVID-19, Kawasaki disease

Cite this article as: Mirkarimi T, Salek M. COVID-19 associated Kawasaki-like multisystem inflammatory syndrome in an adult female with oral, ophthalmic and cardiac manifestation, a case report. Iranian Jour Emerg Med. 2023; 10(1): e16. <https://doi.org/10.22037/ijem.v10i1.41658>.

1. Introduction

COVID-19 can lead to multi-organ dysfunction in severe cases of the disease, often following the release of a large amount of cytokine, known as the cytokine storm (1). During the COVID-19 pandemic, there was a significant increase in the incidence of a Kawasaki-like disease in children, which is referred to as MIS-C.(2, 3) This phenomenon rarely occurs in adults (1). The present case is one of the rare cases of MIS-A with oral, ophthalmic, and cardiac involvement.

***Corresponding Author:** Talayeh Mirkarimi MD; Emergency Medicine department; Imam Ali Hospital, Alborz University of Medical Science, Taleghani Blvd, Karaj, Iran. Phone Number: 00989304665465, Email: t_mirkarimi@yahoo.com, ORCIDs: 0000-0002-8816-8144.

2. Case presentation

A 41-year-old female patient without any underlying diseases presented to the emergency department complaining of bleeding from the gums and red and watery eyes from two days before. The patient had also suffered from persistent high fever, sore throat, weakness, and lethargy from a week before. She did not mention respiratory symptoms such as cough and shortness of breath. She mentioned that several members of her family had contracted COVID-19 over the last two weeks.

The presenting vital signs of the patient were as follows: pulse rate: 110/min, respiratory rate: 18/min, blood pressure: 81/53 millimeter of mercury (mm Hg), oral temperature: 39.4 degrees Celsius (°C), oxygen saturation (spO₂): 95%. There was no evidence of respiratory distress. In the examination, strawberry tongue (Figure 1), cracked lips and gingivitis (Figure 2), bilateral non-purulent conjunctivitis with watery eyes (Figure 3), Redness and slight swelling of both tonsils were

evident. No evidence of lymphadenopathy was observed in the neck. Crackles were auscultated sporadically in the auscultation of the lungs. In the examination of the limbs, symmetrical edema was observed without evidence of peeling in the fingers. No evidence of rash was observed on the patient's skin.

Due to the fever and hypotension, the patient was treated initially with 1-liter intravenous normal saline, and the broad-spectrum antibiotic ceftazidime, which caused a temporary increase in the patient's blood pressure. In the obtained electrocardiogram, sinus tachycardia was observed without significant ST-T changes (Figure 4). In the chest computed tomography (CT) scan, scattered, subpleural, ground-glass involvement was observed in favor of COVID-19 (Figure 5). The initial laboratory results were as follows; white blood cells (WBC): 3600/ μ L, lymphocyte: 540/ μ L, and platelet: 95000/ μ L, erythrocyte sedimentation rate (ESR): 72 mm, and C-reactive protein (CRP): 88 mg/dL, initial cardiac troponin I (cTnI): 0.2 ng/dL, aspartate aminotransferase (AST): 79 mg/dL and alanine transaminase (ALT): 83 mg/dL, lactate dehydrogenase (LDH): 782 mg/dL. Also, the level of troponin increased after six hours in the second test: 0.315 ng/dL. The patient's COVID-19 RT-PCR test was positive.

Due to the hypotension and high level of cardiac troponin, a cardiology consultation was performed, which reported a decrease in the ejection fraction (EF) (40-45%), global hypokinesia, and mild mitral regurgitation (MR) and moderate aortic stenosis (AS) as incidental congenital findings. The patient was admitted to the intensive care unit (ICU) for further treatments. The patient was treated daily with 325 mg of aspirin and heparin with a therapeutic dose. Due to the possibility of a MIS-A diagnosis, the patient was a candidate for intravenous immunoglobulin (IVIG) administration, which could not be procured. The patient was prescribed dexamethasone (8 mg every 8 hours) and Remdesivir according to the national protocol (200mg as a single dose on day 1 followed by 100 mg once a day for the next 5 days). Norepinephrine infusion was initiated due to the patient's hypotension. On the fourth day of hospitalization, the need for norepinephrine was resolved. Due to the absence of a specific infectious source, the antibiotic was discontinued. Patient's gingivitis and conjunctivitis started to get better on the third day and resolved on the ninth and tenth days of hospitalization. Strawberry tongue started to get better on the fourth day but did not resolve completely upon discharge. The dose of dexamethasone was reduced from the fifth day. The patient was transferred to the internal ward on the seventh day of hospitalization. In the echocardiogram performed on the tenth day of hospitalization, global hypokinesia was resolved and EF reached 50-55%. The patient was discharged on the 12th day of hospitalization with good general health. In the 6-month follow-up, the patient lives nor-



Figure 1: The oral manifestation of COVID-19 associated MIS-A as strawberry tongue in the presented patient



Figure 2: The oral manifestation of COVID-19 associated MIS-A as gingivitis in the presented patient



Figure 3: ophthalmic manifestations of COVID-19 associated MIS-A as bilateral conjunctivitis and epiphora

mally without complications.

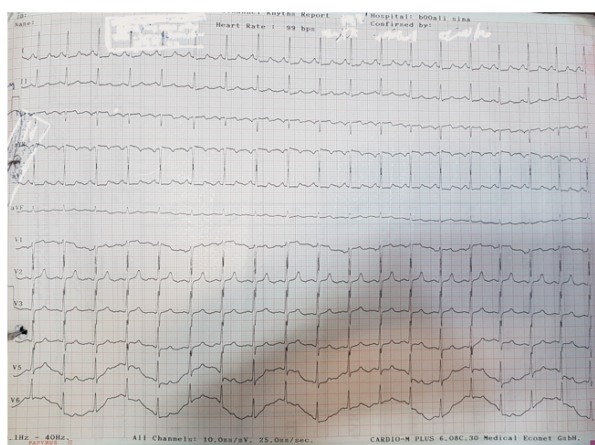


Figure 4: ECG of the patient with sinus tachycardia and no specific ST-T changes

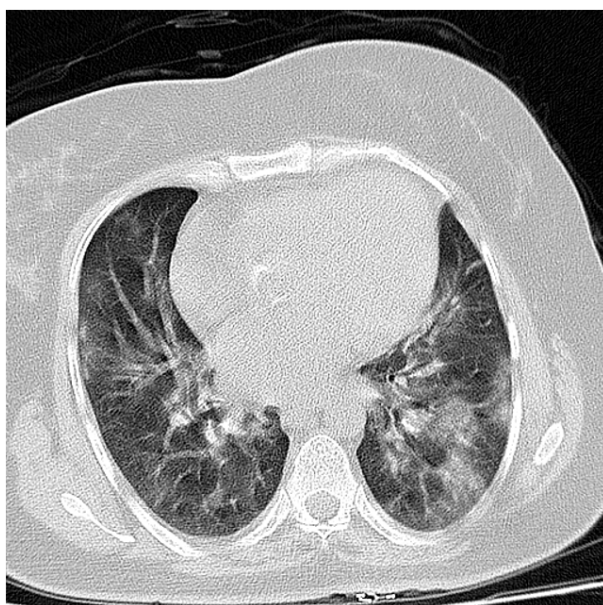


Figure 5: The scattered, sub-pleural, ground-glass involvements was observed in the chest CT scan in favor of COVID-19

3. Discussion

In the COVID-19 pandemic, post-COVID-19 MIS was first defined in children with clinical manifestations of fever and systemic inflammation. The main cause of MIS remains unknown; however, an abnormal response of the immune system to COVID-19 is the most significant theory proposed (4). According to the latest definition of MIS-A offered by the center for disease control and prevention (CDC), the clinical criteria include a fever of more than 38 degrees lasting over 24 hours and at least one of the two criteria including (a) a recent severe cardiac disorder or (b) a rash and bilateral non-purulent conjunctivitis, plus at least two of the follow-

ing criteria: (a) a recent neurological disorder, (b) shock or hypotension, (c) gastrointestinal symptoms, (d) or thrombocytopenia (platelets less than 150,000), and also the presence of laboratory evidence in favor of inflammation and recent infection with COVID-19 (5). The diagnostic criteria of MIS-A were completely met in the presented patient.

In articles related to MIS-A, 63% of the patients have been male and have had no underlying diseases, and 47% of them have been diagnosed with COVID-19. The most common symptoms found in these patients included fever (86%), hypotension and decreased EF (64%), and tachycardia (61%); gastrointestinal symptoms have been less common (6). In many cases of known MIS-A, there have been a 4-week interval between contracting COVID-19 and the presentation of MIS-A symptoms.(7) Treatment of MIS-A have several dimensions which should be considered due to patients' condition such as: supportive care like intubation and ventilation as needed, intravenous fluid therapy, inotropes, anti-coagulant agents, and immunosuppressive medications.(8) In the presented patient symptoms occurred at the same time as contracting COVID-19. Considering the possibility of sepsis or septic shock at the beginning of the patient's visit, it seemed reasonable to prescribe an empiric broad-spectrum antibiotic (9). In the literature reporting MIS-A, various treatments have been considered for these patients. Corticosteroids (e.g., methylprednisolone, prednisolone, and hydrocortisone) in variable doses and durations were the core treatment in most of the reported articles. In these studies, 56% of patients have received corticosteroids and 44% received IVIG.(6) Administering IVIG as part of the treatment has been controversial in articles related to MIS-C.(10) Nonetheless, some articles have shown the effectiveness of IVIG in the recovery of adults.(4, 11) Delphi members suggest IVIG as the first line therapy in MIS-A, particularly in severe forms, in the presence of toxic shock syndrome, or in coronary artery involvement. The recommended dose of IVIG is 2g/kg in a single or divided doses based on the severity of the disease and cardiac abnormality.(12) Tocilizumab and Anakinra (Interleukin-6 (IL-6) and Interleukin-1 (IL-1) receptor antagonists) been used as immunomodulators in less than 5 percent of cases because of their high cost and limitations in provision. Colchicine which has multiple anti-inflammatory mechanisms could be effective in the improvement of cardiac output in patients with cardiogenic shock in MIS-A.(8). In a systematic review, the mortality of MIS-A was reported as 7% (7).

4. Conclusion

Although various treatments have been mentioned for MIS-A in literature, the definite treatment remains uncertain. Identifying MIS-A related to COVID-19 is important for early

treatment which could be effective in reducing mortality and morbidity in these patients. The investigation of the effect of vaccination on the prevalence of this phenomenon requires more extensive studies. The presence of strawberry tongue in the present case was rarely mentioned in the literature in adult cases.

5. Declarations

5.1. Acknowledgments

we thank Dr Mohammad Kaveh, internist who participated in the case management.

5.2. Authors' contributions

Conception and design: TM; Analysing and interpretation of data: TM, MS; Drafting the article or revisiting for important intellectual content: TM, Final approval of the version to be published: TM, MS.

5.3. Conflict of interest

The authors declare that they have no conflict of interest.

5.4. Funding and supports

The authors received no financial support for authorship or publication of this article.

5.5. Ethical consideration and patient consent

Written informed consent was obtained from the patient for publication of this case report. The proposal has been approved by the Research Ethics Committee of Alborz University of Medical Sciences with approval ID: IR.ABZUMS.REC.1401.342 which could be found at: <http://ethics.research.ac.ir/IR.ABZUMS.REC.1401.342>

5.6. Data Availability Statement

All the data was restored safely in Booali Sina hospital, Qazvin province.

5.7. Using artificial intelligence catboats statement

we declare that we did not use artificial intelligence chabots.

References

1. Elouardi Y, Rebahi H, Zarrouki Y, Ziadi A, Younous S, Samkaoui M. COVID-19 associated Kawasaki-like multisystem inflammatory syndrome in an adult. *Revista Española de Anestesiología y Reanimación (English Edition)*. 2022;69(1):43-7.
2. Pouletty M, Borocco C, Ouldali N, Caseris M, Basmaci R, Lachaume N, et al. Paediatric multisystem inflammatory syndrome temporally associated with SARS-CoV-2 mimicking Kawasaki disease (Kawa-COVID-19): a multicentre cohort. *Annals of the rheumatic diseases*. 2020;79(8):999-1006.
3. Alizargar J. The novel coronavirus (COVID-19) and the risk of Kawasaki disease in children. *Journal of the Formosan Medical Association*. 2020;119(11):1713.
4. Shaigany S, Gnirke M, Guttman A, Chong H, Meehan S, Raabe V, et al. An adult with Kawasaki-like multisystem inflammatory syndrome associated with COVID-19. *The Lancet*. 2020;396(10246):e8-e10.
5. Prevention CfDCa. Multisystem Inflammatory Syndrome in Adults (MIS-A) Case Definition and Information for Healthcare Providers [Available from: <https://www.cdc.gov/mis/mis-a/hcp.html>.
6. Behzadi F, Ulloa NA, Danckers M. Multisystem inflammatory syndrome in adults: A case report and review of the literature. *Journal of Medical Case Reports*. 2022;16(1):1-20.
7. 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 network open*. 2021;4(9):e2126456-e.
8. Worku D. Multisystem Inflammatory Syndrome in Adults (MIS-A) and SARS-CoV2: An Evolving Relationship. *BioMed*. 2023;3(1):195-201.
9. Hennon TR, Penque MD, Abdul-Aziz R, Alibrahim OS, McGreevy MB, Prout AJ, et al. COVID-19 associated multisystem inflammatory syndrome in children (MIS-C) guidelines; a Western New York approach. Elsevier; 2020. p. 101232.
10. McArdle AJ, Cunningham AJ, Levin M. Therapy for Multisystem Inflammatory Syndrome in Children. *Reply. The New England journal of medicine*. 2021.
11. Sokolovsky S, Soni P, Hoffman T, Kahn P, Scheers-Masters J. COVID-19 associated Kawasaki-like multisystem inflammatory disease in an adult. *The American journal of emergency medicine*. 2021;39:253. e1-. e2.
12. Hookham L, Fisher C, Manson JJ, Morgan M, O'Hara G, Riley P, et al. Understanding the diagnosis and management of multisystem inflammatory syndrome in adults (MIS-A) in the UK: results of a national Delphi process. *Clinical Medicine*. 2022;22(3):266.