CLINICAL VIGNETTE

The first case of pediatric inflammatory multisystem syndrome temporally associated with SARS-CoV-2 infection (PIMS-TS) in Poland, complicated by giant coronary artery aneurysms

Paweł Tracewski[1], Kamila M. Ludwikowska[2], Leszek Szenborn[2], Jacek Kusa[1]

1 Department of Pediatric Cardiology, Regional Specialist Hospital in Wrocław, Research and Development Center, Wrocław, Poland
2 Department of Pediatric Infectious Diseases, Wrocław Medical University, Wrocław, Poland

On May 3, 2020, a 2-year-old boy in good condition, with no significant medical history, was admitted to our hospital on the 10th day of fever (body temperature of up to 40 °C) after ineffective oral empiric antibiotic therapy with amoxicillin with clavulanic acid. During the first 2 days of the disease, he complained of joint pain and mild diarrhea. No other signs or symptoms were reported. About 5 weeks earlier (at the beginning of the coronavirus disease 2019 [COVID-19] pandemic in Poland), he and his family suffered from mild upper respiratory tract infection accompanied by low-grade fever and rhinosinusitis. No tests were performed at that time.

On admission, differential diagnosis did not indicate any typical cause of fever of unknown origin. The polymerase chain reaction (PCR) nasopharyngeal swab test for severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) yielded negative results twice. The result of the Anti-SARS-CoV-2 ELISA test (Euroimmun, Lübeck, Germany) was positive for immunoglobulin G, and negative for immunoglobulin M.

Laboratory test results are presented in Supplementary material, *Table S1*. No abnormalities were found on chest X-ray and abdominal ultrasound. Transthoracic echocardiography showed a left main coronary artery aneurysm of 4.3 mm in diameter (*z* score, 5.67), a left anterior descending artery aneurysm of 6.3 mm in diameter (*z* score, 13.17), and a right coronary artery aneurysm of 5.4 mm in diameter (*z* score, 5.67) (FIGURE 1), without any myocardial contractility disorders or pericardial effusion.

Except for prolonged fever, none of the typical Kawasaki disease symptoms were present. However, because of laboratory findings and coronary artery aneurysms (CAAs), Kawasaki disease–specific treatment was administered. Fever resolved after intravenous immunoglobulin administration (2 g/kg of body weight). In addition, an antiplatelet agent (acetylsalicylic acid) and warfarin were introduced. The patient was discharged home in good condition. The coronary artery aneurysms persisted on follow-up echocardiography performed after 3 months.

The presented patient was, to our knowledge, the first case of pediatric inflammatory multisystem syndrome temporally associated with SARS-CoV-2 infection (PIMS-TS) in Poland. At that time, except for the Royal College of Pediatrics and Child Health recommendations, no information on this disease was available in the medical literature. Since then, several definitions of PIMS (called multisystem inflammatory syndrome in children [MIS-C] in the United States) were developed.[1-3]

Reports from various countries[1,4,5] have provided a growing set of data on children presenting with signs and symptoms overlapping with Kawasaki disease and toxic shock syndrome, with frequent cardiac involvement and progression to shock. Compared with those data, our patient was at younger age than the median age of patients with Kawasaki disease–like

Correspondence to:
Jacek Kusa, PhD, Department of Pediatric Cardiology, Regional Specialist Hospital in Wrocław, Research and Development Center, ul. Kamieńskiego 73A, 51-124 Wrocław, Poland, phone: +48 71 327 01 40, email: jkusa@poczta.onet.pl
Received: July 3, 2020.
Revision accepted: September 20, 2020.
Published online: September 25, 2020.
Kardiol Pol. 2020; 78 (10): 1064-1065
doi:10.33963/KP.15623

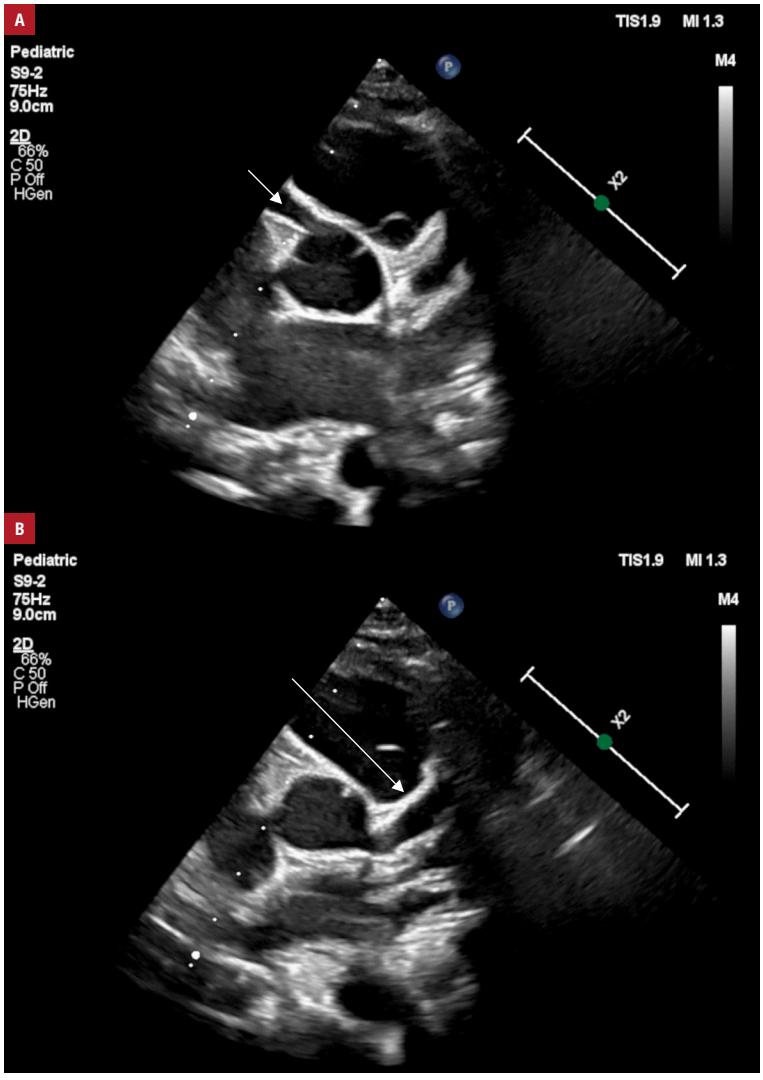

FIGURE 1 Echocardiography in a 2-year-old boy with pediatric inflammatory multisystem syndrome accompanied by giant coronary artery aneurysms: **A** – right coronary artery aneurysm (arrow); **B** – left coronary artery aneurysm (arrow)

PIMS-TS and the presentation was more typical of Kawasaki disease. His disease course was not affected by most commonly described cardiac complications, ie, myocarditis and acute heart failure, but he developed CAAs—another common complication of PIMS. The size of CAAs with a *z* score greater than 10 was observed in the minority of PIMS-TS cases.[5] His clinical symptoms did not correspond with the severity of CAAs, but no clinical or laboratory CAA risk factors have been found so far.[5] Laboratory test results (Supplementary material, *Table S1*) shared a few features with those most commonly observed in PIMS: high levels of inflammatory markers, anemia, lymphopenia, and low sodium concentration. Interleukin 6 levels were not tested, but other biomarkers related to inflammatory response—C-reactive protein concentration and erythrocyte sedimentation rates—were significantly increased.

Unlike most PIMS cases, ferritin and N-terminal fragment of the prohormone brain natriuretic peptide levels in our patient were within the reference range.

Coronary artery aneurysms can complicate all phenotypes of PIMS-TS, not only the Kawasaki disease–like presentation. Our case supports the conclusion derived from the current data, namely, indicating that all children in whom PIMS-TS is suspected should undergo cardiac evaluation. Further research is needed to establish best therapeutic options for these patients, as they may differ from those known in Kawasaki disease before the COVID-19 pandemic.[5]

SUPPLEMENTARY MATERIAL

Supplementary material is available at www.mp.pl/kardiologiapolska.

ARTICLE INFORMATION

CONFLICT OF INTEREST None declared.

HOW TO CITE Tracewski P, Ludwikowska KM, Szenborn L, Kusa J. The first case of pediatric inflammatory multisystem syndrome temporally associated with SARS-CoV-2 infection (PIMS-TS) in Poland, complicated by giant coronary artery aneurysms. Kardiol Pol. 2020; 78: 1064-1065. doi:10.33963/KP.15623

REFERENCES

1 European Centre for Disease Prevention and Control. Paediatric inflammatory multisystem syndrome and SARS-CoV-2 infection in children. https://www.ecdc.europa.eu/sites/default/files/documents/covid-19-risk-assessment-paediatric-inflammatory-multisystem-syndrome-15-May-2020.pdf. Accessed May 15, 2020.

2 World Health Organization. Multisystem inflammatory syndrome in children and adolescents with COVID-19: scientific brief. https://www.who.int/publications-detail/multisystem-inflammatory-syndrome-in-children-and-adolescents-with-covid-19. Accessed May 15, 2020.

3 CDC Centers for Disease Control and Prevention. Multisystem inflammatory syndrome in children (MIS-C) associated with coronavirus disease 2019 (COVID-19). https://emergency.cdc.gov/han/2020/han00432.asp. Distributed via the CDC Health Alert Network on May 14, 2020.

4 Belhadjer Z, Méot M, Bajolle F, et al. Acute heart failure in multisystem inflammatory syndrome in children (MIS-C) in the context of global SARS-CoV-2 pandemic. Circulation. 2020 May 17. [Epub ahead of print].

5 Whittaker E, Bamford A, Kenny J, et al. Clinical characteristics of 58 children with a pediatric inflammatory multisystem syndrome temporally associated with SARS-CoV-2. JAMA. 2020; 324: 259-269.