Takotsubo cardiomyopathy in a toddler with pulmonary atresia and ventricular septal defect

Kenichi Tetsuhara , Shota Muraji, Mamoru Muraoka, Shunsuke Fujii

Fukuoka Children's Hospital, Fukuoka, Japan

Correspondence to
Dr Kenichi Tetsuhara;
ken-tetsuhara@mti.biglobe.ne.jp

Accepted 18 June 2022

DESCRIPTION

A young boy with an 11-month-old history of Rastelli procedure and unifocalisation for pulmonary atresia with ventricular septal defect and major aortopulmonary collateral artery presented to our emergency department because of ill appearance and diaphoresis. The patient had a fever and exhibited the signs of poor feeding 2 days before presentation. On presentation, respiratory failure, shock and comatose were noted. Echocardiography revealed slightly decreased apical and posterior wall motion compared with the data obtained 7 months ago. ECG showed no remarkable changes from the previously obtained data (figure 1A). The results of the laboratory examination were as follows: troponin T level, 0.52 ng/mL; creatine kinase level, 254 U/L; and brain natriuretic peptide level, 419.0 pg/mL. Contrast-enhanced CT showed no anomaly or contrast defects of the coronary arteries. We suspected septic shock because of the patient's fever and shock. Mechanical ventilation and the administration of fluid bolus, norepinephrine, dobutamine and antibiotics were initiated. Subsequently, the patient's circulatory status improved.

Slow ventricular tachycardia occurred on the day following admission (figure 1B), and apical wall motion was further reduced (video 1). The patient was subsequently diagnosed with Takotsubo cardiomyopathy (TC) based on transient regional left ventricular hypokinesia that extended beyond a single epicardial vessel distribution, presence of sepsis as a physical trigger, abnormality noted on ECG, and elevated levels of troponin T and brain natriuretic peptide, according to the diagnostic criteria for paediatric TC.[1] Although mexiletine infusion partially succeeded in controlling ventricular arrhythmia, his blood pressure decreased because of recurrent arrhythmia. The blood pressure increased with the administration of dobutamine. We suspected acute myocarditis as a differential diagnosis; thus, we planned to transfer the patient to another centre for extracorporeal membrane oxygenation. After several hours, his arrhythmia disappeared. Wall motion improved after a week (video 2). His blood, urine and cerebrospinal fluid cultures were negative. The patient was extubated after 7 days; he was discharged 19 days after admission without any morbidity.

TC is typically triggered by stress and is characterised by transient left or right ventricle

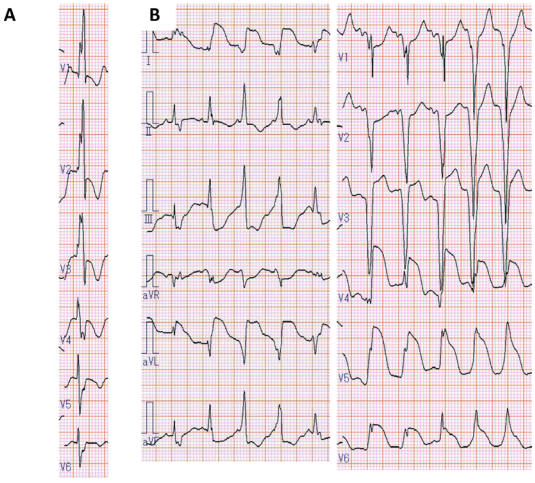
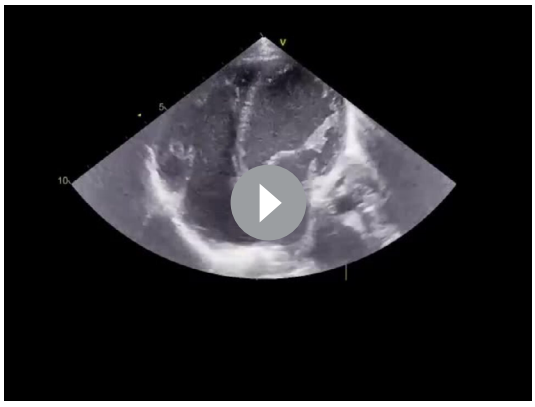
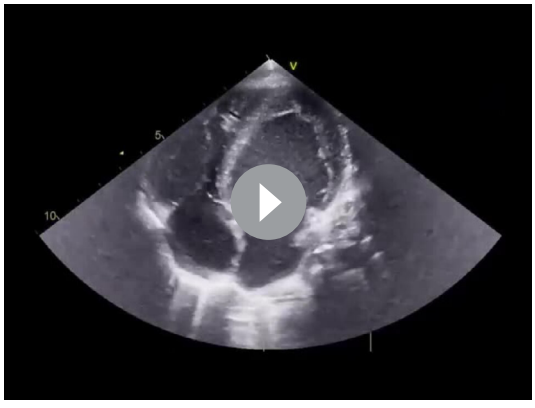


Figure 1 ECG on admission (A) and the day following admission (B). The latter shows slow ventricular tachycardia.



Video 1 Echocardiography on the day following admission shows reduced left ventricular apical wall motion.



Video 2 Echocardiography 7 days after admission shows improved wall motion.

To cite: Tetsuhara K, Muraji S, Muraoka M, *et al*. *BMJ Case Rep* 2022;**15**:e251323. doi:10.1136/bcr-2022-251323

dysfunction with various wall motion abnormalities (typically apical left ventricular wall motion).[1] It is rare in children, particularly young children.[2] Very few cases of TC development in children with congenital heart diseases (CHDs) have been reported.[3 4] To the best of our knowledge, the present case is the first report on TC development in a child with pulmonary atresia and ventricular septal defect. Although changes on ECG in children with TC are reported,[1] arrhythmia has not been indicated, as per our knowledge. Myocardial hypoxia and surgery for CHD may affect stress response. Although TC is often triggered by sepsis in severely ill adults,[5] insufficient information is available for children.[2] TC triggered by sepsis and septic cardiomyopathy partially overlap.[6] Therefore, future studies are warranted to determine the association of TC with CHD and sepsis in children. Thus, when children with CHD who are suspected of having any infection present with decreased cardiac function, TC may be a possible cause in addition to myocarditis and septic cardiomyopathy.

Patient's perspective

The patient's mother said, "I wish I had noticed something wrong with this boy a little earlier."

Learning points

- When children with congenital heart diseases who are suspected of having any infection present with decreased cardiac function, Takotsubo cardiomyopathy may be a possible cause in addition to myocarditis and septic cardiomyopathy.
- Children with Takotsubo cardiomyopathy can develop arrhythmia.

Correction notice This article has been corrected since it was first published online. In the introduction '11-year-old' has been changed to '11-month-old'.

Acknowledgements We thank Dr Suzu Imamura for treatment of the patient and Dr Keiichiro Mizuno for critical advice on the manuscript.

Contributors KT drafted the manuscript, and SM, MM and SF revised it. All authors approved the final version of the manuscript.

Funding The authors have not declared a specific grant for this research from any funding agency in the public, commercial or not-for-profit sectors.

Competing interests None declared.

Patient consent for publication Parental/guardian consent obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

Case reports provide a valuable learning resource for the scientific community and can indicate areas of interest for future research. They should not be used in isolation to guide treatment choices or public health policy.

ORCID iD
Kenichi Tetsuhara http://orcid.org/0000-0001-6473-0326

REFERENCES

1 Topal Y, Topal H, Doğan C, *et al*. Takotsubo (stress) cardiomyopathy in childhood. *Eur J Pediatr* 2020;179:619–25.
2 Sendi P, Martinez P, Chegondi M, *et al*. Takotsubo cardiomyopathy in children. *Cardiol Young* 2020;30:1711–5.
3 Watanabe M, Shiraishi S, Takahashi M, *et al*. Fontan operation in a paediatric patient with a history of takotsubo cardiomyopathy. *Interact Cardiovasc Thorac Surg* 2014;19:326–8.
4 Dalla Pozza R, Lehner A, Ulrich S, *et al*. Takotsubo cardiomyopathy complicating percutaneous pulmonary valve implantation in a child. *World J Pediatr Congenit Heart Surg* 2020;11:NP37–40.
5 Park J-H, Kang S-J, Song J-K, *et al*. Left ventricular apical ballooning due to severe physical stress in patients admitted to the medical ICU. *Chest* 2005;128:296–302.
6 Vallabhajosyula S, Deshmukh AJ, Kashani K, *et al*. Tako-Tsubo cardiomyopathy in severe sepsis: nationwide trends, predictors, and outcomes. *J Am Heart Assoc* 2018;7:e009160.

Correction: *Takotsubo cardiomyopathy in a toddler with pulmonary atresia and ventricular septal defect*

Tetsuhara K, Muraji S, Muraoka M, *et al*. Takotsubo cardiomyopathy in a toddler with pulmonary atresia and ventricular septal defect. *BMJ Case Rep* 2022;15:e251323.

This article has been corrected since it was first published online. In the introduction '11-year-old' has been changed to '11-month-old'.

Case reports provide a valuable learning resource for the scientific community and can indicate areas of interest for future research. They should not be used in isolation to guide treatment choices or public health policy.

BMJ Case Rep 2022;**15**:e251323corr1. doi:10.1136/bcr-2022-251323corr1