Cardiology in the Young

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Being exposed to the same trigger increases the risk of recurrence of takotsubo cardiomyopathy in children and can have fatal results: a case report

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Brief Report

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Abstract

Takotsubo cardiomyopathy has an incidence of 1% of acute coronary syndrome in the adult population, and the risk of recurrence is approximately 1.5% per year. However, only a few cases have been reported in children. Having a neurologic disorder and being exposed to the same trigger repeatedly have been associated with an increased risk.

Introduction

Takotsubo cardiomyopathy is a well-defined disease in adults and is increasingly reported in children. It is characterised by a transitory apical ventricular dysfunction, typically reversible, without evidence of coronary artery obstruction.

Case report

An eleven-year-old boy was admitted to PICU for an epileptic status. He was full-term, born in good condition. He was under regular follow-up by the Neurology Team given his hypotony, epilepsy, and leukoencephalopathy of an unknown aetiology. He was also affected with an ostium secundum atrial septal defect, which was surgically corrected when he was four. During the transfer from the local hospital, the patient was intubated due to the low level of consciousness. He was haemodynamically unstable requiring high doses of inotropes (epinephrine 0.2 mcgr/kg/min and norepinephrine 0.15 mcgr/kg/min). At the admission in our centre, a chest X-ray was performed, which showed acute pulmonary oedema. The transthoracic echocardiogram showed severe left ventricular dysfunction and apical and mid-wall dyskinesia with preserved contractile function of the basal segments (Supplementary Figure S1, Supplementary Video S1).

These findings suggested takotsubo cardiomyopathy. The electrocardiogram showed T-wave inversion in the left precordial leads, and cardiac biomarkers (Troponin I and Pro-brain natriuretic peptide) were highly elevated (maximum of 8196 ng/L and 31,701 pg/ml respectively, on the second day of admission). Regular echocardiograms were performed, and the patient showed an improvement of the left ventricular function. Inotropes were discontinued on the 6th day with an echocardiogram showing a normal left ventricular function, but remaining apical dyskinesia. Other diagnostic tests showed normal thyroid function and negative viral serologies for CMV, EBV, HSV-1, HSV-2, HSV-6, parvovirus, adenovirus, influenza A, and enterovirus.

The patient was discharged on the 10th day on carvedilol. Posteriorly, he was under regular follow-up in the Paediatric Cardiology outpatient clinic. Given the persistence of apical dyskinesia, an MRI was performed, showing the absence of late gadolinium enhancement. The contractility of the apical segments was recovered after one month of the event.

Six months later, the patient had a seizure at home with a cardiorespiratory arrest that required advanced resuscitation for forty minutes. During the transfer to the ICU, the patient had refractory hypotension which required intravenous perfusion of epinephrine with a maximum dose of 0.2mcg/kg/min.

Initially, the echocardiogram showed normal ventricular function. However and given the fact that he required high vasoactive support, the echocardiogram was repeated after twelve hours of admission showing severe ventricular dysfunction with akinesia of the apex and mid-wall of the left ventricle.

Repeated echocardiography was performed daily. Systolic function gradually improved until complete recovery on the tenth day of the event. The electrocardiograms revealed ST elevation and negative T waves in V5-V6. Furthermore, the laboratory analysis found an elevation of Troponin I until 592ng/L and Pro-brain natriuretic peptide until 22.836 pg/ml at admission.

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From the neurological point of view, the patient required three drugs to control the seizures. The brain MRI performed on the 6th day of admission indicated an hypoxic ischaemic injury, with the involvement of bilateral lenticular basal ganglia, thalamus, cerebellum, as well as corticoid subcortical affectation in border territories. These findings were probably related to the cardiorespiratory arrest and low cardiac output. The restriction at the globus pallidus was remarkable, which together with subthalamic and periaqueductal involvement suggested an underlying metabolic disease (probably mitochondrial disease).

Due to a severe neurological impairment, an agreement was reached with the family to withdraw treatment, and the patient died on the tenth day of hospitalisation.

Takotsubo syndrome is characterised by reversible segmental ventricular failure, without involvement of a specific vascular territory. In its classic form, there is apical hypokinesia, although other segments may also be affected. The incidence is similar between boys and girls, in contrast to the adult population in which it is more common in women. The clinical manifestations may vary from asymptomatic presentation to acute heart failure. In paediatrics, it is most often asymptomatic.[1,2]

In most cases, it is associated with physical or emotional stress, which acts as a trigger for the development of cardiomyopathy. Cases related to infections, post-surgical context, neurological insult, or drug abuse have been well described in the literature.[3–6]

The risk of recurrence in children is not well known, as few cases are described, while in adults, it is about 1.5% per year.[7] Psychiatric and neurologic disorders have been observed in association with a higher risk of recurrence in adults.[8] Also, in sporadic case reports, the reiteration of the same trigger has been associated with recurrence as it is shown in the case described.[9] Furthermore, the index and recurrent event showed the same ballooning pattern, which has been described as the most common form of recurrence.[8]

The main treatment of this disease is support therapy. Complete recovery of ventricular systolic function, ranging from a few days to several months, is necessary to confirm the diagnosis.[3] The exact associated mortality rate in children is unknown given the lack of literature. Although the prognosis is favourable in most cases, some case series have described a mortality rate in paediatrics of 7%, associated with anoxic injury predominantly.

In the case report described, it is unclear whether the cardiomyopathy caused the anoxic injury or it was the cardiac arrest or the probable mitochondrial disease.

Further studies are needed for a better understanding of the outcomes and risk of recurrence of takotsubo cardiomyopathy in children.

Supplementary material. The supplementary material for this article can be found at https://doi.org/10.1017/S1047951123001518.

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