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Stress cardiomyopathy following pericardectomy in a child: A case report

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Abstract:
Takotsubo (stress) cardiomyopathy is an acute reversible cause of transient left ventricular dysfunction. It is a diagnosis of exclusion and is exceptionally rare in the paediatric post-operative setting. We report a 10-year-old male who developed refractory hypotension and severe left ventricular dysfunction. This occurred within 24 hours of radical pericardectomy for tuberculous pericardial effusion. Thus, data shows the importance of early recognition of stress cardiomyopathy in unexplained post-operative ventricular dysfunction.

Keywords: Stress cardiomyopathy, takotsubo syndrome, pericardial effusion, pericardectomy, paediatric cardiac surgery, tuberculous pericarditis

Background:
Takotsubo (stress) cardiomyopathy is an acute and reversible cause of transient left ventricular dysfunction. It is a diagnosis of exclusion and requires consideration of alternative causes of acute ventricular dysfunction, including myocarditis and coronary abnormalities such as anomalous left coronary artery from the pulmonary artery (ALCAPA) **[1]**. It accounts for approximately 1–2% of patients presenting with suspected acute coronary syndrome in adults **[2]**. Stress cardiomyopathy is uncommon in children, although paediatric cases have been reported after major physical stress, trauma and cardiac surgery **[3, 4, 12, 19 and 20]**. Physical stress, major surgery and critical illness are recognized triggers of Takotsubo syndrome **[5, 6]**. Excessive catecholamine-mediated myocardial stunning is an important proposed mechanism, with regional differences in sympathetic innervation and β-adrenoceptor signalling contributing to the characteristic wall-motion patterns **[7]**. The InterTAK Diagnostic Criteria provide a standardized framework for diagnosis **[8]**, although their validation in children is limited. Early recognition is important because ventricular dysfunction is usually reversible with supportive management **[9–11]**. Post-operative cardiac surgical triggers have been described in children, including after mitral valve surgery **[3, 12]**. Adrenergic agents used post-operatively may also contribute to myocardial stunning in susceptible patients **[13, 14]**. No universally accepted treatment guideline currently exists **[15]**. Pericardial decompression and pericardiocentesis have also been reported as temporal triggers of stress cardiomyopathy, including in tuberculous pericardial effusion **[20, 21]**. A previous adult case described biventricular stress cardiomyopathy after pericardiectomy for tuberculous constrictive pericarditis **[19]**. Therefore, it is of interest to evaluate a child who developed stress cardiomyopathy following radical pericardectomy.

Case report:
Presentation and initial workup:
A 10-year-old male child presented with a 30-day history of intermittent fever, progressive lethargy and generalised oedema. He was febrile, tachycardic, tachypnoeic and hypotensive, with reduced oxygen saturation on room air. Examination revealed bilateral pitting pedal and periorbital oedema, raised jugular venous pressure and muffled heart sounds. Investigations showed elevated inflammatory markers (CRP 48 mg/L, ESR 78 mm/hr), lymphocytosis and normal serum troponin I. Chest radiograph showed cardiomegaly with a globular cardiac silhouette. ECG showed low-voltage complexes with diffuse ST elevation. Echocardiography confirmed massive organised pericardial effusion with preserved LVEF (60%). Tamponade physiology was evidenced by diastolic right ventricular collapse and respiratory variation in mitral inflow.

Surgical management:
Percutaneous pigtail catheter drainage failed owing to the organised, fibrinous nature of the effusion. Given clinical deterioration and persistent sepsis, surgical drainage was performed under general anaesthesia. Dense caseous pericardial material necessitated conversion from a subxiphoid approach to median sternotomy. Radical pericardectomy and complete debridement were performed. Both phrenic nerves were preserved and closure was performed with pleural and pericardial drains.

Post-operative course and diagnosis:
Immediate post-operative echocardiography confirmed normal biventricular function (LVEF 58%). The child was transferred to the paediatric intensive care unit on low-dose dopamine (5 µg/kg/min). Within 8–12 hours, progressive refractory hypotension developed, unresponsive to fluid resuscitation. Repeat ECG showed new diffuse T-wave inversions and a prolonged corrected QT interval. Echocardiography at 24 hours demonstrated markedly reduced LVEF (20%) with apical and mid-ventricular akinesis and basal hyperkinesis. Mild right ventricular dysfunction was also noted. Serum troponin I was mildly elevated and coronary artery anatomy was normal on echocardiography. Sepsis screening, serum cortisol and thyroid function tests were within normal limits. The clinical picture was consistent with stress cardiomyopathy and met the clinical framework of the InterTAK Diagnostic Criteria [8], while acknowledging that these criteria are not specifically validated in children.

Management and recovery:
Inotropic and vasopressor support was escalated over 72 hours. Drugs used included dopamine 20 µg/kg/min and dobutamine 20 µg/kg/min. Adrenaline 0.2 µg/kg/min and noradrenaline 0.2 µg/kg/min were also administered. Mechanical ventilation was continued with fluid restriction, electrolyte management and prophylactic anticoagulation. LVEF recovered to 40% by 96 hours, permitting gradual inotrope weaning. The child was extubated on post-operative day 5 and LVEF normalised completely (58%) by post-operative day 10. Histopathology confirmed tuberculous pericarditis with caseating granulomas and Langhans giant cells. Anti-tubercular therapy was commenced once haemodynamic stability was achieved. The child was discharged on post-operative day 10 and remained well at 6-week follow-up with normal echocardiography.

Table 1: Differential diagnosis of acute post-operative ventricular dysfunction in children

Diagnosis	Key Features	Distinguishing Investigation	Reversibility
Stress Cardiomyopathy	Apical ballooning, post-stress onset	Echo pattern, normal coronaries	Complete, days–weeks
Myocarditis	Fever, viral prodrome	Cardiac MRI, biopsy	Variable
Septic Cardiomyopathy	Sepsis criteria, bacteraemia	Blood cultures, procalcitonin	Variable
Coronary Artery Disease	Regional wall motion abnormality	Coronary angiography	Depends on injury
Hypothyroidism	Bradycardia, myxoedema	Thyroid function tests	Slow, with treatment

*Sources: Adapted from Ghadri et al. **[2]**, Wittstein et al. **[7]** and Prasad et al. **[17]**.*

Discussion:
Post-operative Takotsubo syndrome in children is exceptionally rare, but published paediatric cases demonstrate that major physical stress and cardiac surgery can precipitate transient ventricular dysfunction **[3, 4, 12, 19 and 20]**. A previous adult report described biventricular stress cardiomyopathy after pericardiectomy for tuberculous constrictive pericarditis, while another report described transient left ventricular dysfunction after therapeutic pericardiocentesis for tuberculous pericardial effusion **[19, 20]**. These reports support the plausibility of pericardial disease and surgical or decompressive stress as triggers, but the present case differs in age and clinical setting. Massive pericardial effusion with cardiac tamponade produces substantial physiological stress and radical pericardectomy may further alter loading conditions and sympathetic activity during the post-operative period **[6, 16 and 19]**. Tuberculous pericarditis adds a chronic inflammatory and infectious stressor to the acute surgical insult. Excessive catecholamine-mediated myocardial stunning remains an important proposed mechanism and may be amplified by surgical trauma, haemodynamic redistribution following relief of tamponade and critical illness **[7]**. The differential diagnosis includes myocarditis, septic cardiomyopathy, coronary abnormalities and endocrine disorders. Distinguishing features are summarised in **Table 1 [17]**. Diagnosis in this patient was supported by the characteristic temporal sequence, echocardiographic wall-motion pattern, mild troponin elevation, normal coronary anatomy and exclusion of major alternative diagnoses. Complete recovery of ventricular function was consistent with the reversible nature of stress cardiomyopathy **[8]**. Paediatric triggers remain uncommon and are largely described in case reports and small series **[9–12, 18]**. Based on the literature identified for this report, we did not find a previous paediatric case specifically describing stress cardiomyopathy after radical pericardectomy for tuberculous pericardial effusion with tamponade; however, this observation should be regarded as a case-level contribution rather than proof of an entirely unprecedented mechanism. Complete ventricular recovery has been reported in many paediatric cases, although outcomes depend on the severity of haemodynamic compromise and associated illness **[9–11]**.

Conclusion:
Stress cardiomyopathy is a rare complication of pericardectomy in children. Early recognition of the characteristic ventricular wall-motion pattern and systematic exclusion of alternative causes are important in unexplained post-operative ventricular dysfunction. Ventricular function can recover substantially with appropriate supportive management, as demonstrated by complete recovery in this case.

Advancement of knowledge:
Novel multi-stressor trigger:
This is the latest documented paediatric case of stress cardiomyopathy following radical pericardectomy for tuberculous effusion with tamponade. This novel trigger-combining a chronic infectious substrate with acute surgical decompression-has not been described before.

Severity does not predict irreversibility:
Complete haemodynamic recovery was achieved despite a nadir LVEF of 20% and multi-agent inotropic support. This reinforces that severity of initial dysfunction does not predict irreversibility in paediatric stress cardiomyopathy.

Diagnostic utility in children:
The InterTAK Diagnostic Criteria were reliably applied in the paediatric post-operative setting, demonstrating their utility for systematic diagnosis and exclusion of alternative aetiologies in children. Taken together, these findings expand the documented spectrum of physiological triggers for paediatric Takotsubo syndrome. They support maintaining a high index of suspicion for this reversible diagnosis in children with unexplained ventricular dysfunction.

Key messages:
[1] Stress cardiomyopathy is a rare but usually reversible cause of severe ventricular dysfunction in the paediatric post-operative setting.

[2] Pericardectomy for tuberculous effusion with tamponade constitutes multiple converging adrenergic stressors capable of triggering myocardial stunning.
[3] Diagnosis requires systematic exclusion of alternative aetiologies and identification of the characteristic apical ballooning echocardiographic pattern.
[4] In this case, prompt supportive treatment was followed by complete ventricular recovery.

Patient perspective:
The patient's family was comprehensively counselled regarding the diagnosis, course and prognosis. Written informed consent was provided for publication.

Author contributions:
[1] Rajat Kalra: Conceptualisation, surgical management, manuscript drafting.
[2] Mukesh Kumar Jaswant Singh Pachahara: Corresponding author, anaesthesia management, critical care, supervision, critical manuscript revision.
[3] Mayank Tanwar: Critical care, post-operative care, data collection.
[4] Hirday Kumar: Anaesthesia management and guidance, critical care, manuscript review.
[5] Gautam Shankar: Paediatric cardiology input, initial diagnosis, echocardiographic interpretation.
[6] Ankita Kumari: Data collection, critical care, anaesthesia management.
[7] Gautam Siddharth: Manuscript writing and editing, OT care.
[8] All authors approved the final manuscript.

Ethics statement:
This case report was conducted in accordance with institutional ethical standards. Formal ethics committee approval was waived as this report describes a single patient. Written informed consent was obtained from the patient's legal guardian prior to inclusion.

Conflicts of interest:
The authors declare no conflicts of interest. No funding was received for this work.

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