Canadian Journal of Cardiology 35 (2019) 544.e3–544.e5 www.onlinecjc.ca

Case Report

Neonatal Cardiac Arrest From Left Ventricular Cardiac Hemangioma: A Surprising Presentation

Valérie Delisle,[a] Jean Perron, MD,[a] Valérie Lafrenière-Bessi, PhD,[a] Jean-Marc Côté, MD,[b] Christian Drolet, MD,[b] Christian Couture, MD, MSc,[c] and Frédéric Jacques, MD, MSc[a]

[a] *Services de Chirurgie Cardiovasculaire Pédiatrique, Québec-Université Laval, Quebec City, Quebec, Canada*

[b] *Cardiologie Pédiatrique, CHU de Québec-Université Laval, Quebec City, Quebec, Canada*

[c] *d'Anatomopathologie et Cytologie, Institut Universitaire de Cardiologie et Pneumologie de Québec-Université Laval, Quebec City, Quebec, Canada*

ABSTRACT

Cardiac hemangioma is rare, even more when leading to a cardiovascular collapse in a seemingly healthy newborn. A 6-day-old neonate had a tamponade caused by a basolateral hemangioma of the left ventricle. Partial surgical resection was performed. A congenital lobular capillary hemangioma was diagnosed upon histologic examination. The patient recovered completely and shows normal development at the 12-month follow-up.

RÉSUMÉ

Les hémangiomes cardiaques sont rares, en particulier ceux qui entraînent un collapsus cardiovasculaire chez un nouveau-né qui semble en santé. Un nouveau-né de 6 jours a présenté une tamponnade causée par un hémangiome basolatéral du ventricule gauche. Une résection chirurgicale partielle a été réalisée. Un hémangiome capillaire lobulaire congénital a été diagnostiqué par examen histologique. Le patient a récupéré complètement et présentait un développement normal au suivi de 12 mois.

Sudden collapse in a seemingly healthy neonate is a devastating event that remains with no causative diagnosis in half of cases.[1] We report the successful management of an unusual left posterobasal ventricular cardiac tumour as a cause of cardiovascular collapse.

Case

A healthy 2-day-old full-term male neonate, normal Apgar score, was readmitted 12 hours following hospital discharge. Fetal ultrasound examinations and perinatal development were unremarkable. Consciousness alteration, colour change, atonia, and subsequent cardiovascular collapse forced immediate cardiopulmonary resuscitation maneuvres by the medically trained father. Vital parameters were recovered upon readmission. The neonate had metabolic acidosis with increased lactates and hypothermia. Transthoracic echocardiography showed normal left ventricular ejection fraction and mild mitral insufficiency. Moderate circumferential pericardial effusion with thickened visceral pericardium and a slightly dyskinetic mass at the basal lateral wall of the left ventricle leaning close to the mitral valve were also noted (Fig. 1A). Chest contrast-enhanced magnetic resonance imaging (MRI) showed an aneurysm with normal myocardial consistency (Fig. 1B). Subsequent echocardiography suggested coronary fistulas originating from the aneurysm and showed increased pericardial effusion. Cardiovascular collapse in a neonate with the combination of a cardiac tumour and pericardial effusion was considered as a surgical indication to explore the pericardium, perform a biopsy of the tumour, and remove it if possible.

Cardiac surgery with cardiopulmonary bypass and Del Nido cardioplegia was performed at 6 days of life. The pericardium was found to be distended, with dark brown-coloured blood. Systemic heparinization was followed by a bright blush in the pericardial cavity. A solid multilobed mass overhung by 2 other nodules was unveiled in the left atrioventricular groove. A hole on the thin surface of the mass was actively bleeding within the pericardium. The hole was controlled, and the tumour was partially resected; the area was reinforced using autologous pericardium. The close proximity of the tumour to the circumflex artery in the left atrioventricular groove prevented complete surgical resection. Intraoperatively, there was no demonstration of communication with any cardiac chamber.

Intraoperative pathological examination revealed that the left ventricular aneurysm was consistent with congenital lobular capillary hemangioma. Permanent histological sections on formalin-fixed paraffin-embedded tissue confirmed the

Received for publication November 29, 2018. Accepted January 23, 2019.

Corresponding author: Dr Jean Perron, Service of Cardiac Surgery IUCPQ, 2725, chemin Sainte-Foy, Quebec G4V 1G5, Canada. Tel.: +-418-656-4717.

E-mail: jean-perron@fmed.ulaval.ca

See page 544.e5 for disclosure information.

https://doi.org/10.1016/j.cjca.2019.01.023

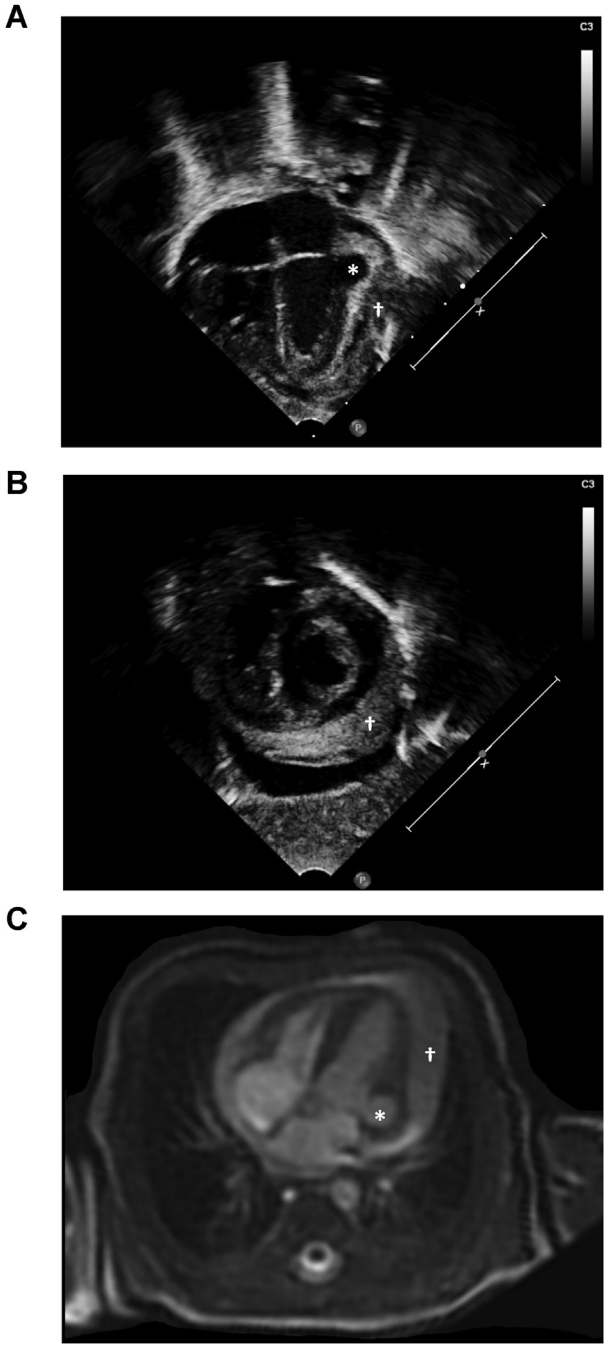

Figure 1. Tumour and pleural effusion imaging. (**A**) 4-chamber echocardiogram showing the dyskinetic tumour (**asterisk**) and the organized pericardial effusion (**dagger**). (**B**) Short axis view showing organized (**dagger**) and liquid pericardial effusion. (**C**) Four-chamber cardiac magnetic resonance imaging showing the dyskinetic tumor (**asterisk**) and the pericardial effusion (**dagger**).

diagnosis, which was supported by immunohistochemical reactivity of tumour cells with endothelial marker CD34 and absence of reactivity for GLUT-1 (Fig. 2).

Genetic investigations for metabolic causes were inconclusive, although the infant had abnormal levels of select amino acids. Of possible interest, the mother had polycystic ovarian syndrome with hereditary tyrosinemia, whereas the

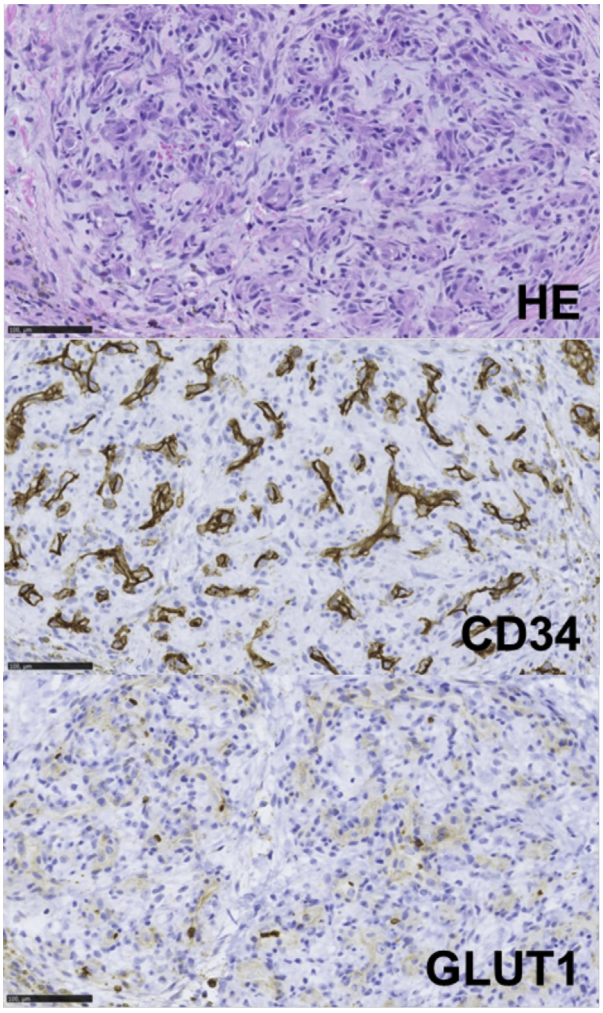

Figure 2. Histology. On hematoxylin-eosin (HE) sections, the tumour consists of lobules of bland endothelial cells without necrosis, atypia, or mitoses. Tumour cells express endothelial immunohistochemical marker CD34 but not GLUT1. Micrometric scale 100 μm.

father's genetic analysis was negative for IVS12+5G−>A in the FAH gene, the main genetic mutation responsible for the autosomal recessive disease of tyrosinemia.

The infant was discharged at postoperative day 19, with normal biventricular function. At 1 year of life, follow-up echocardiogram showed normal heart function with the presence of a residual hemangioma and no signs of thrombus or coronary fistulas. The child remains symptom free, with normal development.

Discussion

This report highlights the successful management of a rare case of neonatal cardiovascular collapse that is usually with no apparent cause but with high rates of fatal or debilitating outcomes.[1] The clinical impact of early diagnosis of fetal or neonatal cardiac tumour could help prevent an unforeseen life-threatening event.

In the current case, prompt resuscitation and surgical management gave favourable outcomes. The clinical presentation of hemodynamically significant tamponade in this case

likely resulted from spontaneous intrapericardial rupture of the tumour. This hypothesis is supported by the fact that the patient developed active intrapericardial bleeding when systemically heparinized for bypass in the operating room. An alternative explanation could be myocardial ischemia and secondary ventricular arrhythmia, as the tumour and the circumflex artery were closely related. The pericardial effusion could then be the consequence of the trauma of cardiac resuscitation.

The presence of the tumour at birth and the absence of reactivity for GLUT-1 in tumour cells are both associated with congenital hemangioma, a type of hemangioma not associated with spontaneous involution. Infantile hemangiomas, on the other hand, are usually not present at birth and regress spontaneously.[2] Despite the benign nature for cardiac hemangiomas, cardiovascular complications are reported.[3-5] Li et al., in a review of 200 cases of cardiac hemangioma (comprising 7% fetuses and another 8% before the age of 10), noted that sudden death is the presentation in 3.5% of the time.[5] Most of these events were related to pericardial effusion and tamponade. Myocardial ischemia was also reported. Surgical indications remain controversial, but removal is recommended in symptomatic patients. Complete removal is preferable to partial resection and biopsy, as it leads to better long-term outcomes.[5] In the case reported herein, the procedure was initially diagnostic and hemostatic. Even if planned, complete removal would not have been possible, as injury to a major coronary artery within the atrioventricular groove would be difficult to repair and a disastrous event in a neonate.

Disclosures

The authors have no conflicts of interest to disclose.

References

1. Herlenius E, Kuhn P. Sudden unexpected postnatal collapse of newborn infants: a review of Cases, Definitions, Risks, and Preventive Measures International Classification of Disease. Transl Stroke Res 2013;2:236-47.

2. Van Vugt LJ, van der Vleuten CJM, Flucke U, Blokx WAM. The utility of GLUT1 as a diagnostic marker in cutaneous vascular anomalies: a review of literature and recommendations for daily practice. Pathol Res Pract 2017;213:591-7.

3. Djordjevic SA, Kalanj J, Intensive N, Unit C. Brief report cardiac haemangioma associated with a duct-dependent congenital heart disease in a newborn infant. Cardiol Young 2017;27:990-2.

4. Sharma J, Hirata Y, Mosca RS. Surgical repair in neonatal life of cardiac haemangiomas diagnosed prenatally. Cardiol Young 2009;19:403-6.

5. Li W, Teng P, Xu H, Ma L, Ni Y. Cardiac hemangioma: a comprehensive analysis of 200 cases. Ann Thorac Surg 2015;99:2246-52.