



Thrombus formation in the heart following balloon atrial septostomy in transposition of great arteries

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Abstract

Thrombotic complications following balloon atrial septostomy (BAS) are unusual. We report a patient with thrombus formation at the site of BAS, extending into the inferior vena cava (IVC), following BAS for transposition of great arteries with intact ventricular septum (TGA-IVS). An urgent arterial switch operation (ASO) with removal of the thrombus was performed.

Keywords Transposition of great arteries · Thrombosis · Balloon atrial septostomy

Introduction

Thrombotic complications following balloon atrial septostomy (BAS) are unusual except thrombus at the peripheral access site [1]. We report a patient who had thrombus formation along the inter-atrial septum that extended into the inferior vena cava (IVC) and hepatic veins early after BAS for transposition of great arteries with intact ventricular septum (TGA-IVS). An urgent arterial switch operation (ASO) with removal of the thrombus was performed.

Case report

A 45-day-old infant weighing 3 kg presented to the emergency department with progressive cyanosis ($\text{SpO}_2 = 40\%$) and worsening of hemodynamics. After resuscitation, transthoracic echocardiogram (TTE) revealed TGA-IVS and a restrictive atrial septal defect (ASD), without additional lesions. The left ventricular (LV) cavity was D-shaped. The LV posterior wall

measured 2.8 mm and the LV mass index measured 30 g/m^2 indicating early regression. Prostaglandin-E1 infusion (0.05 mcg/kg/min) was started. An emergency BAS was planned. The results of the blood gas analysis before and after BAS are shown in Table 1.

Vascular access was through the femoral vein into which a 5-Fr pediatric sheath was placed. As is the practice worldwide [1], 1 mg/kg ($1 \text{ mg} = 100 \text{ units}$) heparin was administered intravenously. A 5-Fr Fogarty catheter was passed via the vascular sheath and through the restrictive ASD into the left atrium (LA). The balloon was inflated with 2 mL 1:1 diluted contrast, and its position in the LA was confirmed under cinefluoroscopic and TTE guidance. The balloon was pulled back three times, and after graduated inflation, a BAS was performed and the adequacy of the septostomy was confirmed by TTE. BAS was uneventful with immediate improvement in systemic saturation (75%) and hemodynamics. Reverse transcriptase polymerase chain reaction test for coronavirus disease (COVID-19) was negative.

As dictated by our previous experience, and those of others [2, 3], we scheduled the patient for an elective ASO with mechanical circulatory assist using extracorporeal membrane oxygenation (ECMO). It was planned to integrate the ECMO circuit in the cardiopulmonary bypass (CPB) circuit [4], as it was clearly anticipated that mechanical circulatory assist would be needed in view of the regressed LV. This has been our preferred strategy over LV retraining with a pulmonary artery band and a systemic-pulmonary artery shunt, which has its own problems [2, 3, 5]. The details of integrating an ECMO circuit in the CPB circuit with its advantages are discussed in a previous paper from our institution [4].

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Table 1 Results of blood gas analysis

Parameter	Before BAS	After BAS
pH	7.25	7.36
pCO ₂	35 mmHg	40 mmHg
pO ₂	18 mmHg	30 mmHg
SPO ₂	40%	75%
HCO ₃ ⁻	10 mmol/L	23 mmol/L
BE _{ecf}	-10 mmol/L	-8 mmol/L
BE _b	-8 mmol/L	-6 mmol/L
Na ⁺	135 mmol/L	136 mmol/L
K ⁺	4 mmol/L	3.8 mmol/L
Lactate	7 mmol/L	2.8 mmol/L

All values are at an FiO₂ of 100%

BAS balloon atrial septostomy

Pre-operative coagulation parameters were normal without evidence of a hypercoagulable state. There was no systemic infection. There were no vascular complications at the vascular access site, there was no discoloration of the lower limb, the arterial pulses were normal, and there was no evidence of deep vein thrombosis (DVT).

Two days following BAS, there was a precipitous fall in systemic saturation with progressive hemodynamic instability. Prompt mechanical ventilatory support was instituted. Repeat TTE revealed a large solitary thrombus restricting blood flow at the BAS site and extending into the IVC and hepatic veins (Fig. 1, Video 1). Instead of an elective procedure, the patient was now taken for emergency removal of the thrombus and ASO.

Operative technique

After sternotomy, a pericardial patch was harvested and was fixed in 0.6% glutaraldehyde solution. Following systemic heparinization, the superior vena cava (SVC) was cannulated using a 14-Fr angled venous cannula and routine CPB was established. A 14-Fr angled venous cannula pointing downwards was chosen to drain the right atrium (RA) completely. Care was taken that the purse-string suture for the IVC was

placed slightly higher than the RA-IVC junction to avoid dislodging the thrombus. Even after placing the canula higher up, adequate venous return was still achieved, and the RA was completely drained. The systemic flow was 450 mL at 35 °C. At moderate hypothermia (28 °C), we achieved a systemic flow of 300 mL till up to cooling to 18 °C [prior to deep hypothermia and circulatory arrest (DHCA)]. Meanwhile, after cooling to 28 °C, a single dose of del Nido cardioplegia was administered into the aortic root for myocardial protection.

The initial part of the ASO comprising harvesting of the coronary buttons from the aorta, transfer into the neo-aortic root, neo-aortic reconstruction, and proximal right ventricular outflow tract (RVOT) reconstruction was performed during the cooling phase under cardioplegic arrest. After the patient was cooled to a nasopharyngeal temperature of 18 °C, DHCA was performed, the RA was opened, and the IVC canula was removed for complete visualization of the thrombus, which was seen at the site of the BAS and extended into the IVC and hepatic veins (Fig. 2). This thrombus occluded the opening in the interatrial septum, probably explaining desaturation and hemodynamic instability even after successful BAS. All the thrombi were completely removed. Following this, the IVC was washed off all debris and the IVC canula was reinserted into the RA-IVC junction and was pushed down further for better drainage. The site of the BAS was carefully inspected for any abrasions or petechiae etc. that could account for endothelial damage at the site of the interatrial septum. However, these were absent.

Following closure of the ASD, the RA was closed and CPB was resumed. Gradual rewarming was started, and the right ventricular outflow tract was reconstructed to complete the operation. The total circulatory arrest time was 16 min; aortic cross-clamp time was 118 min.

Repeated attempts to wean off from CPB were unsuccessful; hence, CPB time could not be calculated, and mechanical circulatory support was therefore instituted to enable LV preparation.

Adequate LV mass and function were not achieved even after 10 days of ECMO, and multiple attempts at weaning from ECMO were unsuccessful. Sepsis and multiorgan failure ensued, and ECMO was finally terminated on the 10th post-

Fig. 1 Pre-operative transthoracic echocardiography. **A** Apical four-chamber view with thrombus obstructing patent foramen ovale. Left ventricle is regressed. **B** Subcostal view showing thrombus at inferior vena cava causing partial obstruction to blood flow. IVC: inferior vena cava, LA: left atrium, PFO: patent foramen ovale, RA: right atrium



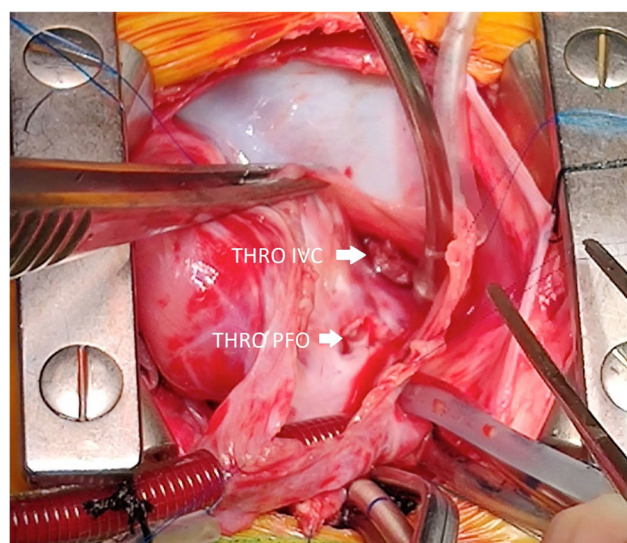


Fig. 2 Intraoperative view showing two separate thrombi at patent foramen ovale and inferior vena cava. IVC: inferior vena cava, PFO: patent foramen ovale, THRO: thrombus

operative day. The family did not consent to an autopsy. Histopathological examination revealed a thrombus that was sterile on aerobic, anaerobic, and fungal cultures.

Discussion

BAS is a percutaneously performed interventional procedure in patients with TGA-IVS to ensure admixture of oxygenated and deoxygenated blood and to achieve hemodynamic stability prior to the definitive ASO. This report focuses on an intracardiac thrombus as a possible complication of BAS. The merits and demerits of ASO in patient with TGA-IVS with regressed LV are not discussed in detail.

Complications following BAS are minimal and are mechanical, traumatic, embolic, electrical [1], rupture of the balloon with fragment embolization [1], inability to deflate the balloon [1], and balloon inflation at an inappropriate site. Traumatic complications are rare with inadvertent injuries to atrial appendages, mitral valve, pulmonary veins, and IVC. Cerebro-vascular accidents such as intracranial bleed and stroke are extremely rare [1].

Patients with surgical documentation of an intracardiac thrombus, which occurred following BAS, have practically never been reported. On an exhaustive search of the literature, we encountered only one brief letter to the editor with a similar diagnosis [6]. However, the patient in that report did not undergo surgery; hence, there was no confirmatory evidence of the pre-operative diagnosis. The authors of that report hypothesized that DVT in their patient was the cause of the thrombus in the RA following BAS.

Theoretically, such a complication may also result from invasive intracardiac lines, inadequate heparinization during

BAS, or secondary infection. However, none of these risk factors were present in our patient before/after entry of the catheter used for BAS. There were no invasive lines in our patient, no signs of any DVT, and no evidence of any infective focus. Intra-operative findings did not suggest trauma to the endocardium or exposure of the myocardium that could have led to thrombus formation, neither were the coagulation parameters deranged before and after BAS. Protein C and S testing, genetic studies, and clotting propensity were not performed as a hypercoagulable state was not anticipated.

An emergency operation to remove the thrombus along with ASO was indicated in our patient. Hence, we did not have an opportunity to evaluate the patient in detail for DVT. Even though DVT was not evaluated objectively by us, there is still a possibility of sub-clinical DVT as the source for the intracardiac thrombus. However, there was no clinically manifest DVT.

Practices of systemic heparinization at our center are like those being followed worldwide [1]. For a simple procedure like BAS, only 100–200 units/kg heparin is administered. In most centers globally, for a simple procedure like BAS at the start or completion, activated clotting time (ACT) is not routinely checked and is presumed to be in the desired range unless there is a prior hypercoagulable state. However, ACT estimation is performed when the procedure is expected to last more than 2 h or at the end of the procedure.

Thrombolysis with tissue plasminogen activator (tPA) to manage the thrombus and stabilize this sick patient prior to surgery was an option. However, we wanted to eliminate the possible risk of stroke due to tPA that could increase significantly if thrombolysis were to be unsuccessful and the patient would have been subjected to CPB for thrombus removal and ASO. It could also be argued that we could have proceeded with an emergency ASO instead of an initial BAS in this critically sick patient, as the LV was not expected to improve with only a BAS. However, we preferred resuscitation and hemodynamic stabilization and BAS as this is our institutional policy. In addition, a BAS before the ASO provided us an opportunity to rule out associated comorbidities and systemic infection.

This report clearly illustrates that even after a successful BAS, a large thrombus did develop at the BAS site. Therefore, if sudden systemic desaturation and hemodynamic deterioration develops following BAS, the cardiologists and surgeons should entertain this diagnosis and appropriate corrective measures to tackle the thrombus should be instituted. These should be tailored on a case-to-case basis.

Supplementary Information The online version contains supplementary material available at <https://doi.org/10.1007/s12055-022-01331-4>.

Author contribution

- 1: ST: first author and operating surgeon
- 2: GK: co-author, chief resident on case
- 3: SR: consultant cardiologist, diagnosis and critical inputs on manuscript
- 4: PG: consultant anesthesiologist, performing preoperative echocardiography and facilitating surgery, critical inputs on manuscript
- 5: SKC: chief of surgery, critical inputs on manuscript

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Data availability Available only on request from the authors as per institutional guidelines due to privacy/ethical restrictions.

Declarations

Statement of human and animal rights All procedures performed in this study were in accordance with the ethical standards of the institutional and national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. No animals were involved in the study.

Informed consent Informed consent was obtained from all individual participants included in the study. The parents of this patient consented to publish this report.

Ethics approval Not applicable.

Conflict of interest None.

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