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ORIGINAL ARTICLE

Routine Cardiac Catheterization Prior to Fontan Operation: Is It a Necessity?

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Abstract

Prior to the Fontan procedure, patients with single ventricle physiology with Glenn shunt are typically referred for cardiac catheterization to assess hemodynamics and potentially provide interventional measures. Currently, echocardiography provides detailed information which together with other non-invasive imaging such as CT scan and MRI may obviate the need for routine cardiac catheterization prior to the Fontan procedure. In this study, we examine the findings in cardiac catheterization in this population to determine: (a) the accuracy of echocardiography in providing adequate information prior to the Fontan procedure, particularly in identifying those in need of per-catheter intervention, and (b) the percentage of patients requiring interventional procedures during cardiac catheterization. We performed a retrospective chart review of echocardiographic and cardiac catheterization data for patients who underwent pre-Fontan cardiac catheterization at our center in the period from 02/01/2008 to 02/28/2017. We aimed to re-examine the necessity of routine cardiac catheterization in all single ventricle patients. This was performed through examining pre-catheterization echocardiography reports and comparing them to findings of the subsequent cardiac catheterization reports. Echocardiography reports were evaluated for accuracy in identifying significant anatomical or hemodynamic findings, which may impact success of Fontan procedure as well as the ability of echocardiography to predict findings important to know prior to the Fontan procedure. In this cohort of 40 children, 3 patients were found to have significant hemodynamic findings through cardiac catheterization which were not previously known by echocardiography. In addition, 28 out of 40 patients (70%) required interventional procedures to address significant abnormalities (systemic to pulmonary arterial collaterals, pulmonary artery stenosis, aortic arch stenosis, etc.). All cases of aortic arch stenosis were detected by echocardiography, however, all patients who required systemic to pulmonary arterial or left SVC embolization were not detected by echocardiography. Furthermore, echocardiography did not detect the need for branch pulmonary artery stenosis in 50% of cases. Cardiac catheterization appears to be an essential part of patient assessment prior to Fontan completion in patients with single ventricle physiology. This current practice may change in the future if a non-invasive screening tool is found to have high positive and negative predictive values in identifying the subset of patients who require potential intervention in pre-Fontan cardiac catheterization.

Keywords Fontan · Pre-Fontan evaluation · Pre-Fontan cardiac catheterization · Routine cardiac catheterization · Echocardiography

Introduction

Single ventricle patients routinely undergo cardiac catheterization as part of pre-Fontan evaluation. Cardiac catheterization provides valuable anatomical and hemodynamic information. In addition, needed per-catheter interventions can be performed during cardiac catheterization to optimize cardiovascular physiology prior to undergoing the Fontan procedure [1, 2].

In the current era with the advent of high resolution echocardiography and other non-invasive angiographic tools, there is a growing interest in avoiding cardiac catheterization in low-risk single ventricle patients [3–5]. The benefits of routine cardiac catheterization in pre-Fontan evaluation should be weighed against the risk of adverse outcome of catheterization in this group which has been reported to be

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approximately 11% based on a recent large multicenter study [1].

In this study, we sought to examine the necessity of an elective cardiac catheterization prior to Fontan completion and to determine if echocardiography and assessment of clinical data can be a predictor of patients in whom cardiac catheterization is not required.

Methods

This is a single-center retrospective study, which was done at Rush University Medical Center. We reviewed medical records of patients who met the inclusion criteria from February 2008 till February 2017.

Inclusion and Exclusion Criteria

We included all patients with single ventricular physiology at Glenn stage that had an elective pre-Fontan cardiac catheterization evaluation.

Exclusion included patients in whom pre-Fontan evaluation echocardiography was performed at another institution or more than 3 months prior to catheterization. In addition, patients with interrupted inferior vena cava were excluded as these patients usually undergo "Kawashima procedure" instead of the classic Glenn procedure.

Echocardiography data were extrapolated from the last echocardiography report preceding cardiac catheterization. Echocardiography was considered complete when all the following were evaluated: single ventricular systolic function, atrioventricular (AV) valve anatomy and the degree of regurgitation (if any presents), single ventricle outflow/s, ascending aorta, aortic arch and isthmus (in patients with risk of coarctation), superior vena cava (SVC) to pulmonary artery (PA) connection site, and pulmonary artery flow pattern.

Cardiac Catheterization

The following data were collected from the cardiac catheterization report: pulmonary vascular resistance (PVR), Glenn circulation mean pressure, single ventricle end-diastolic pressure, transpulmonary gradient, angiographic data, and procedure type if any was performed.

Subsequently, we divided cardiac catheterization into two subgroups:

1. Cardiac catheterization with significant new information or those requiring intervention (significant catheterization): Those include cases where the cardiac catheterization provided new important diagnostic information which was not known prior to the cardiac catheterization, such as elevated Glenn pressure, elevated single ventricle end-diastolic pressure, as well as cases with significant anatomical defects requiring intervening such as presence of PA branch stenosis requiring balloon dilation and or stenting, presence of significant systemic to pulmonary collaterals (SPCs) required embolization, etc.
2. Cardiac catheterization where there were no significant findings or requiring intervention (insignificant catheterization).

Statistical Analysis

Descriptive statistical analysis was performed. Continuous data were reported as mean ± standard deviation (SD) or as median with interquartile range (IQR).

Results

Forty patients met our inclusion criteria (21 males and 19 females). Underlying cardiac anomalies varied with the most common diagnosis being hypoplastic left heart syndrome in 12 (30%) patients. Other anomalies included tricuspid atresia in 10 patients, unbalanced atrioventricular canal in 7 patients, pulmonary atresia in 4 patients, double outlet right ventricle in 4 patients, pulmonary atresia with intact ventricle septum in 2 patients, and one patient with complicated anatomy. Median age at pre-Fontan evaluation was 24 months, IQ range (20.5–31.3) months.

See Table 1 which summarizes demographic and anatomical data.

Among our cohort, echocardiography was complete in 34 patients and was incomplete in 6 patients (4 uncooperative patients and poor acoustic window in 2 patients). None of the echocardiography reports commented on the presence or absence of SPCs.

In 12 patients (30%), echocardiography missed important diagnostic information (6 patients had Left superior vena cava draining to coronary sinus, 4 patients had pulmonary artery stenosis, and 2 patients had narrowed SVC at Glenn anastomosis site). All the above missed anatomical information required transcatheter intervention.

All cardiac catheterization procedures were performed under general anesthesia. Twenty-eight patients (70%) required at least one interventional procedure (coiling of significant SPCs in 16 patients followed by pulmonary artery stenting/angioplasty in 8 patients, aorta angioplasty/stenting in 6 patients, closure of the left superior vena cave in 6 patients, SVC stenting in 2 patients, and balloon dilation of restricted atrial communication in 1 patient). Out of the 28 patients, 10 (35%) patients had two or more procedures during the same pre-Fontan catheterization. See Table 2 for more details.

Table 1 Summary of demographic, clinical, and anatomical variables

Demographic and clinical data	
Sex: male/female (total)	21/19 (40)
Age at Glenn procedure, month (median, IQ range)	5 (4–8)
Age at pre-Fontan evaluation, months (median, IQ range)	24 (20.5–31.3)
Age at Fontan surgery, months (median, IQ range)	27 (20–34)
Weight at pre-Fontan evaluation, kg (median, IQ range)	11.8 (8.1–41.8)
BSA at pre-Fontan evaluation, m^2 (median, IQ range)	0.51 (0.47–0.56)
Anatomical diagnosis	
HLHS	12
TA	10
Unbalanced AVC	7
DORV	4
PA	4
PA/IVS	2
Others	1
Total	40

BSA body surface area, *HLHS* hypoplastic left heart syndrome, *TA* tricuspid atresia, *AVC* atrioventricular canal, *DORV* double outlet right ventricle, *PA* pulmonary atresia, *PA/IVS* pulmonary atresia with intact ventricular septum

Table 2 Procedures done during pre-Fontan evaluation

Intervention type	Number	Percentage (%)
SPCs coiling	16	40
PA angioplasty/stenting	8	20
Aortic angioplasty/stenting	6	15
LSVC to CS coiling	6	15
Others	8	20
Two or more procedures	10	35
Any intervention	28	70

SPCs systemic to pulmonary collaterals, *PA* pulmonary artery, *LSVC* left superior vena cava, *CS* coronary sinus

Table 3 Summary of hemodynamic data which were obtained prior to any transcatheter intervention

Hemodynamic data (mean ± SD)	
Single ventricle diastolic pressure	7.3 ± 2.5 mm Hg
Transpulmonary gradient	4.4 ± 1.5 mm Hg
Glenn mean pressure	11.7 ± 2.3 mm Hg
Calculated PVR	1.7 ± 0.63 wood unit × m^2

PVR pulmonary vascular resistance

Patients in this cohort had normal mean pulmonary vascular resistance and transpulmonary gradient. Glenn circuit mean pressure was normal in 37 patients (92.5%). Hemodynamic data are summarized in Table 3. Three patients had elevated mean Glenn and single ventricle end-diastolic pressures with normal PVR resulting in postponing (2 patients) or exclusion (1 patient) from Fontan operation. The 2 patients (with postponed Fontan surgery) had favorable single ventricle anatomy by echocardiography findings and had no significant lung disease or other comorbidities with impact on proposed surgery. In addition, one more patient was excluded from Fontan completion and subsequently received a heart transplant based on combined echocardiographic and cardiac catheterization findings (moderate AV valve regurgitation and increased Glenn and single ventricle end-diastolic pressures).

Discussion

In 1971 Dr. Fontan and Dr. Baudet [6] performed the first total cavopulmonary connection ‘‘Fontan Procedure’’ which is the final palliative stage for single ventricle patients. Since then, multiple techniques and modifications were introduced. However, the ultimate goal of all different procedures is to achieve near complete separation of pulmonary venous return from the systemic venous return. As such, all systemic venous circulation is directed to the Fontan circulation, except for coronary sinus blood which continues to drain into the common atrium, bypassing the heart [7].

Many investigators believe that routine pre-Fontan cardiac catheterization is a mandatory step to assess the single ventricle hemodynamic data such as pulmonary artery pressure, pulmonary vascular resistance, and single ventricle end-diastolic pressure. In addition, it can detect abnormalities which are needed to be intervened on prior to Fontan completion such as significant SPCs, single ventricle outflow obstruction, and pulmonary artery stenosis. SPCs and

PA stenosis are notorious to be difficult to detect and are frequently missed on conventional echocardiography [3, 8].

In this study, echocardiography failed to recognize PA stenosis in 4 out of the 8 patients (one with poor acoustic window and the rest were not detected despite good echocardiographic studies). Similar findings were reported by Brown and colleagues who found that echocardiography was successful in only 73% of the times to accurately delineate pulmonary artery anatomy [9].

In this cohort, PA balloon angioplasty/stenting was performed in 8 patients (20%). This is comparable to the 16.7% reported by multicenter study published by (C3PO) group [1].

All patients in this study had favorable PVR. This finding is in accordance with Banka's study [3]. Nevertheless, the surgery was postponed in 2 patients and was canceled in 1 other patient based on other hemodynamic findings, such as elevated ventricular end-diastolic pressure and AV valve regurgitation.

Calculated PVR during pre-Fontan evaluation is based on less than full cardiac output—pulmonary blood flow (QP) in patients with Glenn is less than the systemic cardiac output (QS). Therefore, it is impossible to predict what the pulmonary vascular resistance would be after Fontan completion when patients will have increased to full or near full QP [10]. Nonetheless, elevated PVR > 3–4 W.U × m^2 is a predictor for both increased morbidity and mortality in single ventricle patients. Hence, we believe even if a pre-Fontan PVR is unlikely to represent PVR after Fontan completion, accurate calculation of PVR prior to Fontan surgery is crucial for risk stratification and planning for safe timing of total cavopulmonary connection surgery [8, 11]. In addition, cardiac catheterization provides other hemodynamic data (i.e., single ventricle diastolic pressure and Glenn pressure, etc.) which are desired to be within a certain range prior to completing Fontan [12–14].

In this study, we found the most frequent procedure conducted was coiling of SPCs which was done in 16 patients (40%). This finding is close to the stated rate (48%) by the multicenter study reported by the C3PO group [1].

The presence of SPCs in single ventricle may reach more than 80% [15]. The impact of SPCs on clinical outcomes is not fully understood. Many reports believe that SPCs leads to significant energy loss, increased single ventricle end-diastolic pressure, worsening single ventricle function, prolonged pleural effusion and hospitalization duration in the immediate period after Fontan completion [16, 17]. In addition, the presence of significant SPCs would pose a challenge on accurate evaluation of PVR [10, 18]. On the other hand, other investigators such as Bradley et al. [19] believe SPCs have no significant adverse effect on the immediate post-operative outcomes. Due to the poor understanding of the effect of SPCs on short and long outcomes, significant practice variability continues to be present among cardiac centers on how to deal with SPCs [5, 19–22]. As such, no current consensus exists regarding the indications of SPCs coiling. Therefore, occlusion criteria are subjective and lack scientific validity. Examples of these proposed criteria include stepping up in the oxygen saturation in the pulmonary artery when compared with SVC sample, filling defect as a result from negative contrast wash out from the SPCs when pulmonary artery angiography is performed, detecting pulsatility of pulmonary artery in the absence of pulsatile Glenn, elevated single ventricle end-diastolic pressure, and size of the collateral vessels [23]. Despite cardiac magnetic resonance (CMR) may be a reliable non-invasive technique for evaluating SPCs flow [24–26], this method has not been standardized yet.

In children with Glenn, the estimated additional pulmonary blood flow provided by collaterals is 1.59 L/min/m^2 with a range of (0.54–3.34) L/min/m^2 [25]. Dori et al. [27] have demonstrated that SPCs embolization has significant favorable acute hemodynamic outcomes with an approximate decrease in the collaterals flow of an average of 50% and increased systemic blood flow with no drop in the systemic saturation. However, long-term outcome is still unclear leading to controversy and practice inconsistencies to continue.

Cardiac catheterization in pre-Fontan patients was found to be needed in approximately half of patients. In these patients, cardiac catheterizations revealed new diagnostic information affecting perioperative management and/or required essential transcatheter intervention [3]. In our cohort, 70% of our patients had an intervention with or without other significant new diagnostic information resulting in postponement or excluding of Fontan completion. Our interventional rate was in agreement with the overall rate (69%) reported by the centers participating in C3PO project. Interestingly, interventional rate varies significantly among all 8 participating centers of this study with a range of 50–84% [1].

In addition to the well-known possible acute adverse events of cardiac catheterization [1], there is a growing awareness of hazardous long-term effect of ionizing radiation. The risk of radiation-related cancer in patient who received radiation during childhood seems to be related to the total radiation dose and it is more prominent in children irradiated early in life. In addition, the risks for solid tumors persisted throughout life [28]. Due to the above, there is mounting interest to substitute pre-Fontan cardiac catheterization by non-invasive methods [3–5, 29].

Ro and his colleagues [5] had created and tested an algorithm based on clinical and echocardiography findings in order to characterize the subgroup of patients in whom routine pre-Fontan cardiac catheterization can be forgone. Despite they concluded the ability of criteria

to discriminate patients who could skip catheterization (negative predictive value) was high (93%), 3 out of 46 patients (in whom catheterization deemed to be not indicated) received important transcatheter interventional procedure. In addition, they found that the algorithm is inadequate to detect PA stenosis as well as to assess the presence of significant SPCs. Even though adding magnetic resonance imaging to their algorithm would substantially improve its ability to detect PA stenosis and SPCs collaterals, this was not tested in their work and the validity of their algorithm has not been verified by different centers.

Another non-invasive algorithm has been reported recently by Yasin et al. [4]. This algorithm utilized non-invasive angiography modalities such as CMR/or cardiac CT scan in combination with clinical and echocardiographic data. This algorithm was tested retrospectively on 44 patients and it showed a good sensitivity and specificity in detecting adverse post-operative outcomes. Interestingly, the sensitivity of the test did not change after adding hemodynamic data obtained by cardiac catheterization as an additional risk factor. Therefore, they concluded that magnetic resonance imaging and computed tomography can be a good substitute for pre-Fontan cardiac catheterization, which should be reserved for cases in which non-invasive evaluations are not satisfactory or need an intervention. However, the investigators have not reported the percentage of patients who received transcatheter intervention based merely on new data derived by cardiac catheterization (i.e., significant SPCs or PA branch stenosis) which was missed by their algorithm. In addition, to our knowledge the validity of the algorithms neither has been tested prospectively nor validated by different institutions.

Non-invasive angiographies modularity such as CMR allow better delineation of the single ventricle anatomy including the aortic arch, branch pulmonary arteries, indices for pulmonary vascular growth, and significant SPCs in patients with congenital heart disease. Thus, when it's compared with echocardiography, its utilization in a routine pre-Fontan evaluation could more accurately discriminate those patients who will need an intervention and possibly will decrease the need for invasive angiography and therefore decreasing radiation exposure. Despite CMR can provide valuable anatomical information, it is incapable to offer hemodynamic data (i.e., single ventricle end-diastolic pressure) which could be detrimental on mortality and long-term outcomes [24–26, 30–33]. In addition, prospective large studies comparing anatomical data derived by cardiac catheterization versus CMR are deficient and cost effectiveness yet needs to be studied.

Conclusion

In this study, pre-Fontan cardiac catheterization was essential in 28 patients (70%) as it either added new essential diagnostic information and/or provided an opportunity to deliver therapeutic intervention to help in rehabilitating the cardiovascular system. The latter is important to maximize the ideal anatomy and eliminate risk factors such as significant systemic to pulmonary collaterals, pulmonary artery stenosis, etc.

Echocardiography cannot detect SPCs and it frequently missed PA stenosis even among patients in whom echocardiography was deemed to be complete. Till a new non-invasive modality is shown to be reliable in evaluating the single ventricle anatomy and providing accurate hemodynamic information (such as single ventricle pressure, Glenn pressure, and pulmonary vascular resistance), routine cardiac catheterization is a pre-requisite for all patients prior to Fontan operation.

Limitations

Our study is a retrospective study of a single-center experience with small sample size, which prevents conduction of subgroup analysis. In addition, our findings need further verification on a larger scale by conducting a multicenter study.

Due to the lack of universal consensus on when to intervene, our data may be affected by our center bias toward aggressive transcatheter treatment of collateral vessels which is may be the case in many but not all institutions.

Compliance with Ethical Standards

Conflict of interest The authors declare that they have no conflicts of interest related to this study.

Ethical Approval This study was agreed to be conducted by our Institutional Review Board.

Informed Consent Informed Consent was waived as this study was a retrospective chart review study.

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