EDITORIAL COMMENT

Cost Containment in the Single Ventricle Population*

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n this issue of JACC: Advances, O'Byrne et al1 assessed the differences in cumulative hospital costs and cost-per-day-alive between the 2 treatment arms of the SVR (Single Ventricle Reconstruction) trial. The study cohort consisted of 303 patients with single ventricle heart disease from 9 centers in the United States, and costs were compared serially at 1, 3, and 5 years following the Norwood operation. The authors found that the differences in total costs and cost-per-day alive between the modified Blalock-Taussig (mBT) shunt group and the right ventricle-pulmonary artery (RV-PA) conduit group did not reach statistical significance at any of the prespecified follow-up time points. Similar results were observed within the subgroup of transplant-free survivors of the Norwood operation.1

We congratulate the authors in attempting to better characterize the costs associated with staged palliation of single ventricle heart disease using one of the largest available prospective multicenter datasets. One crucial element that impacts hospital costs, postoperative morbidity, and resource utilization in management of single ventricle heart disease is parental socioeconomic status (SES). This was not completely addressed in this study. There is a growing body of evidence that suggests an association between lower SES and adverse outcomes following the Norwood operation, including neurodevelopmental and functional status outcomes at

6 years.²⁻⁴ In the majority of these studies, SES was conceptualized as a composite that includes racial, ethnic, economic, and neighborhood factors that cumulatively represent a family's or individual's relative sociodemographic position based on income, education, and occupation.5 While SES is often challenging to quantify and characterize, various reports, especially those in the single ventricle population, have defined SES using validated metrics, such as the Area Deprivation Index⁶ or Diez-Roux score.⁷ The randomized nature of the SVR trial likely removes any confounding bias related to SES between the 2 treatment arms. However, exclusion of SES and related metrics (including insurance and payer status) from this analysis represents a missed opportunity, especially since socioeconomic disparities have been shown to influence outcomes in the SVR cohort itself.^{2,7} Socioeconomic and demographic factors are of particular importance for patients with single ventricle disease as they often require frequent follow-up and intensive parental involvement and support.8,9 Additionally, SES may play a prominent role when discharging patients after the Norwood operation since ensuring familial resources to participate in home-monitoring programs is essential during the interstage period. 10 Importantly, such programs require diligent parental involvement and convenient access to medical facilities capable of providing emergency pediatric care. However, parents from lower socioeconomic strata may not have the resources requisite for safe discharge following the Norwood operation, and hospitalizations are prolonged and inpatient costs accrue while such services are arranged. This has particular significance for parents that have other dependents at home, as well as caregivers unable to take an extended hiatus from work. The cost implications of SES in the single ventricle disease population thus warrant further investigation.

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Another important factor with particular relevance to clinical practice relates to the subcomponents of hospital charges and costs addressed in this article. Inpatient costs represent a proxy for resource utilization involving various hospital services, including intensive care unit and operating room use, medical supplies, laboratory tests, pointof-care evaluations, pharmacy-related services, radiographic and imaging services, equipment use, and professional services, among others. 11 Dissecting these costs in greater granularity may aid identification of areas of disproportionately higher costs with regards to either mBT shunt or RV-PA conduit use. In turn, these areas may be targeted for tailored cost containment strategies. For instance, data from the SVR trial have previously identified important geographic and center-specific variations in health care utilization and costs following the Norwood operation. Costs related to room and board and laboratory tests were the principal drivers of disparities among centers. While this may be related to center-specific practice, several postoperative complications were found to occur significantly more frequently at high-cost centers, including pleural effusion, seizures, wound infection, thrombus, liver dysfunction, sepsis, and necrotizing enterocolitis.12 These data suggest that cost optimization may be more appropriately guided by the post-Norwood hospital course rather than choice of pulmonary blood flow. Further analyses into subcomponent-specific inhospital and postdischarge costs may thus help contextualize the findings from the present study.

Leveraging the strengths of the SVR trial, the study authors succeeded in the herculean task of characterizing the short- and long-term costs associated with staged palliation of single ventricle heart disease, as well as in elucidating any cost-related differences with regards to shunt strategy. However, the implications remain nebulous, and it is unclear how the findings of this study should inform decision-making. For one, assuming that overall resource utilization, including in-hospital cost, is crudely

associated with postoperative morbidity and the requirement for unplanned reinterventions,13 the primary result of the present study is perhaps not surprising given the findings of the SVR trial¹⁴ and all subsequent follow-up studies.15 Specifically, at 6 years following the Norwood operation, the hazards of death or transplant and catheter interventions were not different between the RV-PA conduit and mBT shunt groups.¹⁶ Furthermore, while O'Byrne et al identified aortic atresia and prematurity as significant predictors of increased cost-per-day-alive across the entirety of the SVR follow-up period, both these preoperative factors are unmodifiable. Mitigation of costs, and more importantly, attrition, may therefore be better achieved with closer postoperative surveillance of high-risk neonates and infants, along with prompt identification and treatment of residual lesions¹⁷⁻¹⁹ rather than with a decision between 2 ostensibly similar sources of pulmonary blood flow. Indeed, clinicians should be encouraged to adopt the surgical strategy that provides the best clinical outcomes for their patients, rather than be influenced by shunt-associated cost variations that did not reach statistical significance in a robust analysis. Nevertheless, O'Byrne et al should be congratulated on an excellent study that opens various avenues for further research, including cost analyses in the single ventricle disease population as they relate to modifiable comorbidities, postoperative nutrition and feeding, successful completion of home-monitoring programs, and long-term qualityof-life metrics.

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