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Transcatheter Management of Coronary Sinus Atresia in an Infant with Severe Cardiac Dysfunction Following Superior Cavopulmonary Anastomosis for Univentricular Palliation

Sok-Leng Kang, MBBS & Lee N. Benson, MD

Abstract

Coronary sinus ostial atresia (CSOA) associated with coronary venous drainage through a persistent left superior vena cava is rare. However, preoperative recognition of this anomaly is vital in infants undergoing cardiac surgery that might inadvertently lead to impairment of coronary venous return. We report transcatheter treatment of CSOA in an infant with severe cardiac dysfunction following superior bidirectional cavopulmonary anastomosis. Successful decompression of the atretic coronary sinus with radiofrequency perforation and successive balloon dilation resulted in improved ventricular function. Transcatheter management of CSOA is feasible and an attractive alternative to surgery, particularly in patients with significant single ventricle dysfunction.

Keywords: Coronary sinus atresia, functional single ventricle, percutaneous, perforation

Introduction

Coronary sinus ostial atresia (CSOA) is rare and typically associated with anomalous coronary venous drainage through a persistent left superior vena cava (LSVC) and/or intra-cardiac Thebesian veins into the atrium. Although an intrinsically benign anomaly, CSOA associated with a LSVC has serious implication in children with complex congenital heart defects undergoing cardiac surgery.¹ Specifically, creation of a superior bidirectional cavopulmonary connection (BCPC) during second stage univentricular palliation with or without disconnection of the LSVC, can obstruct coronary venous egress resulting in myocardial ischaemia. Surgical strategies to secure coronary venous drainage under such circumstances are well described with good outcomes.²⁻⁵ Here, we describe percutaneous management of CSOA in an infant with single ventricle physiology and severe cardiac dysfunction following a superior BCPC.

Case Report

A 5-month-old infant with double outlet right ventricle, mitral atresia, and bilateral superior vena cava with a bridging innominate vein underwent bidirectional BCPC and Damus-Kaye-Stansel procedure. The right superior vena cava (SVC) was anastomosed to the right pulmonary artery and a very small LSVC was left intact due to intraoperative suspicion of coronary sinus (CS) ostial stenosis. Her previous procedures included pulmonary artery (PA) banding and coarctation repair at four days old,

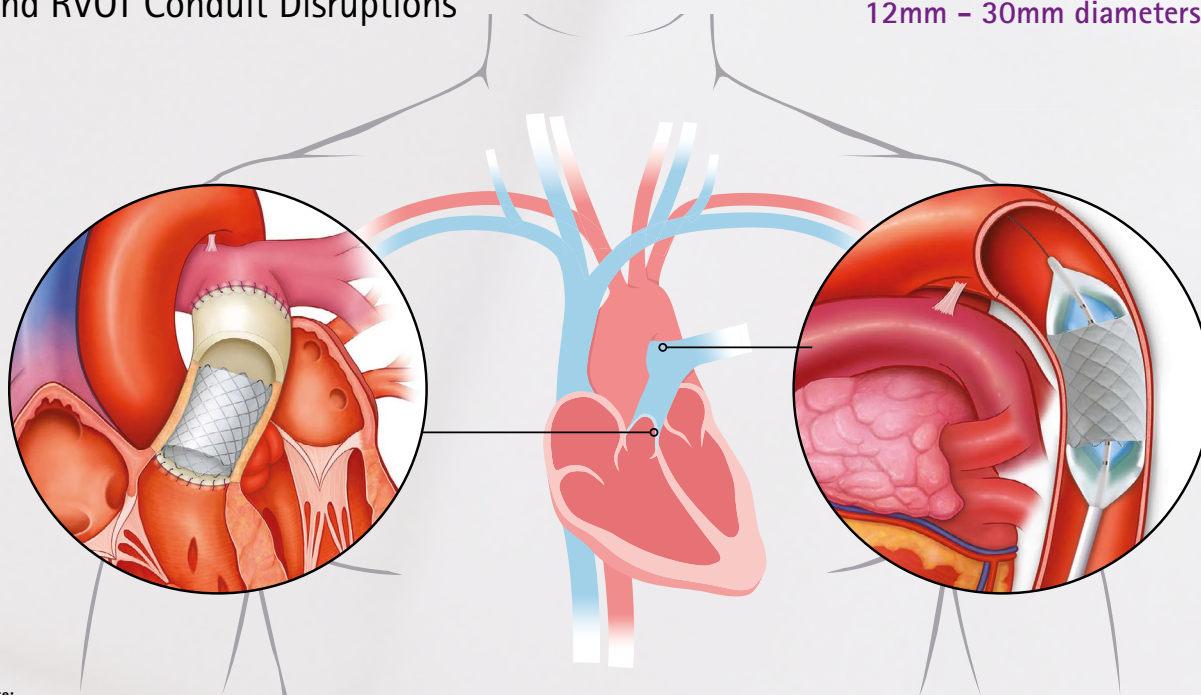
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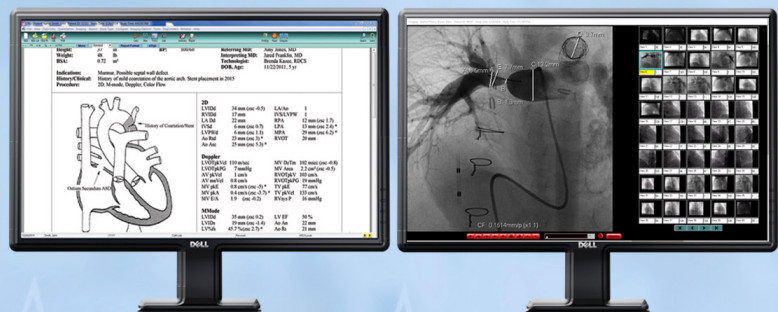
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followed by repeated balloon dilations of the PA band due to progressive cyanosis when she was four months old. On the first post-operative day, the infant had a cardiac arrest shortly after extubation with profound ventricular dysfunction and was cannulated emergently to extracorporeal membrane oxygenation (ECMO). In the absence of cardiac recovery and increasing concerns over adequacy of pulmonary blood flow, the child was taken to the cardiac catheterization laboratory on Day 7 of ECMO support for further evaluation. Initial angiography via the left internal jugular access demonstrated stenosis of the LSVC with decompressing channels to the vertebral plexus and inferior vena cava. The CS was atretic with extensive retrograde filling of the cardiac veins (**Figure 1**). We cannulated the CS with a 5F JR2.5 catheter (Cordis, Santa Clara, CA, USA) and the atretic CS was perforated with a Nykanen wire (Baylis Medical, Montreal, Canada) into the right atrium (RA) with radiofrequency energy of 3W applied for one second. The Nykanen wire was then replaced with a Grand Slam coronary wire, over which sequential dilations of the perforation were performed with 3mm and 5 mm Trek balloons (Abbott Vascular, Illinois, USA). Following decompression of the CS, there was a reduction of CS mean pressure from 13mmHg to 7mmHg and LSVC pressure from 17mmHg to 7mmHg. The right SVC pressure remained at 15mmHg. Repeat angiography in the CS showed rapid contrast entry to the RA and no extravasation of contrast (**Figure 2**). Echocardiography showed gradual recovery of ventricular function in the ensuing 48 hours. Despite improved cardiac function, persistent desaturation made weaning from ECMO support difficult. On Day 12 of ECMO, bedside surgical ligation of the LSVC was performed. On Day 14, a further cardiac catheterization demonstrated an unobstructed BCPC but mildly hypoplastic proximal branch PAs. Mean right SVC pressure was 8mmHg. She underwent balloon dilation of bilateral branch pulmonary arteries and embolization of venovenous collaterals. Subsequent ECMO course was complicated by a massive cerebral infarction and significant coagulopathy, and life-sustaining therapy was ultimately withdrawn on Day 17 of ECMO.

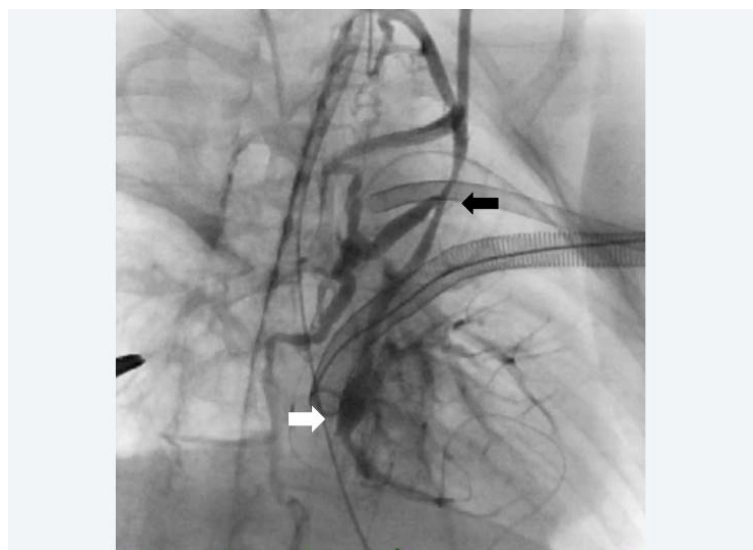


FIGURE 1 Angiogram via the left internal jugular access demonstrating stenosis of the left superior vena cava (black arrow) with decompressing channels to the vertebral plexus and inferior vena cava. The coronary sinus was atretic (white arrow) with extensive retrograde filling of the cardiac veins.

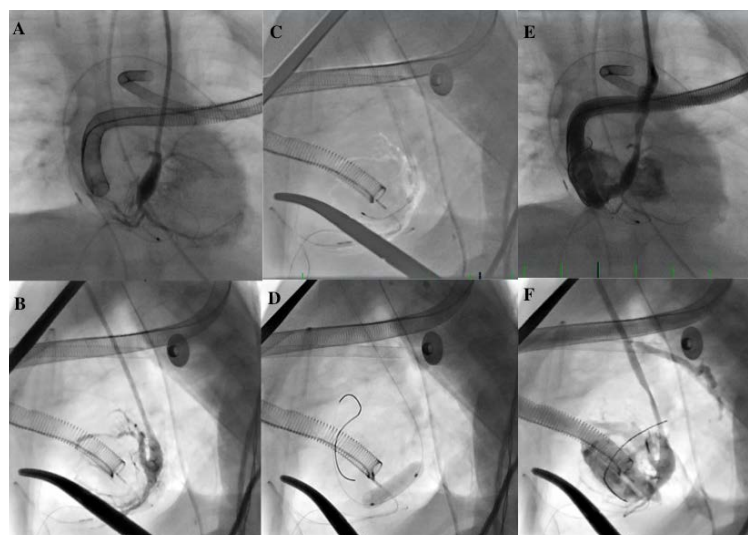


FIGURE 2 (A,B) The atretic coronary sinus (CS) was cannulated with a 5F JR2.5 catheter and the position of the venous ECMO cannula in the right atrium was used as a target for a (C) radiofrequency perforation of the CS with a Nykanen wire. (D) The perforation was dilated sequentially with 3mm and 5mm coronary balloons. (E,F) Repeat angiography showed widely patent CS with rapid contrast entry to the RA.

Discussion

Recognition of CSOA and persistent LSVC is crucial prior to a superior BCPC in staged univentricular repair. In the majority of children with bilateral SVCs, both caval veins are divided and anastomosed to the pulmonary arteries. If the LSVC was considerably smaller, particularly in the presence of a bridging innominate vein, the LSVC is simply ligated to prevent venous runoff to the right atrium. When the LSVC is the only outlet draining an atretic CS, disconnection of the LSVC results in interruption of coronary venous egress, acute coronary venous hypertension and myocardial ischaemia. Even when the LSVC is preserved, the higher-pressure in the caval system after a superior BCPC can adversely affect the coronary arterio-venous gradient and myocardial perfusion.⁶ Our patient had normal ventricular function preoperatively; thus, in retrospect, the cardiac arrest was likely precipitated by progressive coronary venous obstruction due to the stenotic LSVC and elevated venous pressures post BCPC. Further, recovery of cardiac function was observed following decompression of the atretic CS.

When CSOA is diagnosed pre- or intra-operatively in the absence of ventricular dysfunction, surgical treatment has yielded good outcomes. Various surgical techniques of CS unroofing or rerouting of LSVC to the atrium are well reported.¹⁻⁵ Transcatheter management of CSOA offers a less invasive approach and may reduce operative risks particularly in the setting of significant ventricular dysfunction. To the best of our knowledge, percutaneous treatment of CSOA associated with single ventricle physiology has only been reported twice in the literature. El-Said et al described successful radiofrequency perforation of an atretic CS and subsequent stent implantation in a 3-month-old infant with progressive ventricular dysfunction following Norwood Stage 1 procedure.⁷ Petit et al described a similar technique in a young adult who presented with symptomatic heart failure after Fontan completion.⁸ Both showed good



recovery of cardiac function and symptomatic improvement at short-term follow-up.^{7,8} From a technical perspective, the trajectory of CS perforation can be guided by placing a snare or curved tip catheter at the right atrial end of CS; or in our case, using the existing venous ECMO cannula in the RA as a target. Additionally, multimodality imaging such as transoesophageal echocardiography, pre-procedural computed tomography (CT) or magnetic resonance image (MRI) fusion with live fluoroscopy may be useful for procedural guidance. Once unobstructed coronary sinus flow to the RA is established, the LSVC can be occluded percutaneously or ligated surgically at the time of BCPC or Fontan.^{7,8}

The diagnosis of CSOA is challenging, with the majority of reported cases found incidentally during coronary angiography, intra-operatively or at autopsies.^{2,3,9} In infants with complex cardiac anomalies undergoing single or biventricular repair; a thorough knowledge of the anatomy of systemic veins and patency of coronary sinus is necessary to inform surgical planning. The presence of left SVC on preoperative echocardiogram, with retrograde flow and/or dilation of coronary sinus should prompt suspicion of coronary sinus atresia and further evaluation with CT or MRI.⁹ Similarly, when a persistent LSVC is identified during cardiac catheterization, the direction of flow should be scrutinized. If retrograde flow is suspected, selective LSVC angiography is indicated to determine the patency of the coronary sinus and presence of anomalous pulmonary venous drainage.¹⁰

Conclusion

Atresia of the coronary sinus associated with persistent LSVC is rare. However in the setting of univentricular repair, one should always remain cognizant of the possibility of CSOA to avoid the catastrophic consequences of interrupted coronary venous drainage. Our case supports the feasibility and safety of percutaneous decompression of CSOA. The transcatheter approach is less invasive and an attractive alternative to surgery in patients with single ventricle dysfunction.

Declarations

Funding: None

Conflicts of interest: The authors declare that they have no conflict of interest.

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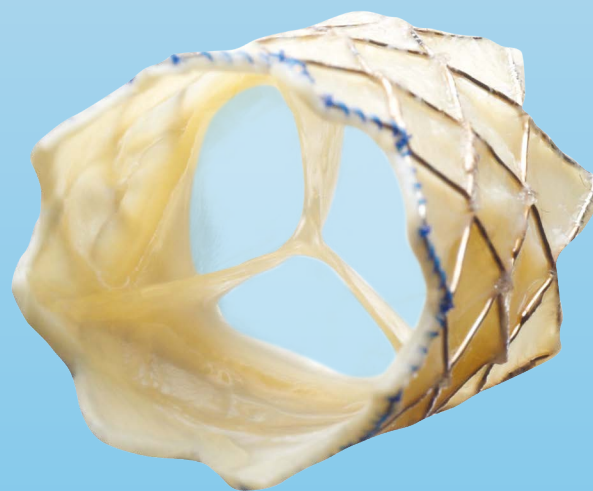
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- The **UH/RBC Heart Center is an internationally recognized program partner with the Nationwide Children's Hospital Heart Center in Columbus, OH collectively forming The Congenital Heart Collaborative (TCHC).** This partnership joins resources in providing the most comprehensive highest quality care on both campuses. TCHC is a dedicated service line with a common executive administration and functions as one program on two campuses with the commitment to expand access to high-quality comprehensive cardiac care regardless of patient age to the communities we serve while equally embracing a scholarly and educational mission. TCHC provides excellent cardiothoracic surgical, cardiac interventional, electrophysiologic, and non-invasive services. An example of the success of our partnership is our fetal cardiac interventional service comprised of members from both campuses and based at UH/RBC.
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- Academic affiliation with Case Western Reserve University School of Medicine.

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defects (e.g. ASD, VSD, PDA, etc), where the connections from the vena cavae-right atrium-right ventricle-pulmonary arteries are normal for the right heart, and left atrium-left ventricle-aorta, for the left heart (atrio-ventricular and ventriculo-arterial concordance and normal levocardia). The codes for abnormal connections are designed to capture the patients with a more complex congenital heart anatomy involving any abnormal atrio-ventricular or ventriculo-arterial connections, such as variations of single ventricle anatomy, transposition, dextrocardia, etc. Importantly, post-surgical connections through shunts/conduits are considered abnormal connections (e.g. Blalock Taussig or Sano shunts, Glenn and Fontan patients, and those with homografts). Soon after these codes pass the RUC process and values are assigned, CICCW has planned for an SCAI webinar to explain the codes in detail and answer any questions providers may have. The webinar should take place in late 2020 and will likely be repeated in late 2021 as a refresher prior to the codes being available for use on January 1, 2022. This further delay adds another layer to the examples provided in **Figure 1**, caused by the printing process, as the 2021 CPT® book has already been printed and soon to be released.

Finally, for the current project at the CPT® application stage, we will be introducing new transcatheter repair of aortic coarctation codes this October, just as this edition of *Congenital Cardiology Today* comes out in print. This application includes three new codes, two for coarctation stenting and one for angioplasty alone. The stent codes are divided into two categories: one for involvement of any major side branches (such as transverse arch stenting or stenting of abdominal aorta for mid-aortic syndrome), and the second code for the more typical juxta-ductal coarctation where no head and neck vessels are involved. For consistency, this model was copied from the one used

for endovascular graft for aortic aneurysms, designed to rightfully capture the increased complexity of stenting the more complex defects. Finally, a code for balloon angioplasty of aortic coarctation has been designed for all scenarios and with the intent of capturing all of the work performed as compared to the peripheral extremity angioplasty code (CPT® 37246) that we have been relegated to for so many years. Please refer to **Table 1** for a timeline of this project as well as future projects.

The CICCW would like to extend a special thanks to the leadership at SCAI for their continued support of this project. Additionally, our gratitude to Dr. Robert Vincent for his contributions to the work of the CICCW. Dr. Vincent has rotated off the workgroup and is being replaced by Dr. John Rhodes.

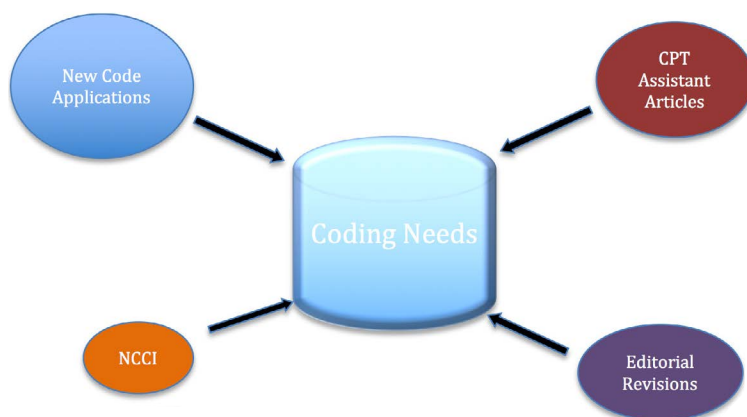


FIGURE 2 Various pathways for resolution towards individual coding needs

TABLE 1 Project Timelines

Project	CPT Meeting Deadline	CPT Meeting Presented	RUC Survey	RUC Meeting Presented	Status	When Available	New code (value)
Pulmonary artery angiography with device closure bundles		June 2017	n/a	n/a	Passed	Dec 2017	n/a
Directed by CMS to create new pericardiocentesis codes		Sep 2018	Nov 2018	Dec 2018	Passed	1-Jan-2020	33016 (4.4), 33017 (4.62), 33018 (5.4)
Modifier 63 for infants under 4 kg		Sep 2018	n/a	n/a	Passed	Jun 2019	n/a
Atrial septostomy	Nov 2019	Feb 2020	Feb 2020	Apr 2020	Passed	1-Jan-2021	33XX0*
Intracardiac stenting	Nov 2019	Feb 2020	Feb 2020	Apr 2020	Passed	1-Jan-2021	33XX1* and 33XX2*
ICE for all use with all cardiac cath procedures		May 2020	n/a	n/a	Passed	1-Jan-2021	
Congenital diagnostic catheterization base codes	Feb 2020	May 2020	Jun 2020	Oct 2020		1-Jan-2022	
Stent / angioplasty for repair of coarctation of the aorta	Jun 2020	Oct 2020	Oct 2020	Jan 2021		1-Jan-2022	
Pulmonary artery ductal stenting	Feb 2021**	May 2021**	Jun 2021**	Oct 2021**	Drafted		
Intra-cardiac / vascular RF perforation					First draft		
Possible Hybrid procedure with co-surgeon code					Discussions		
Completed	* Final code number to be determined, values approved and soon to be released						
In process / scheduled	** Anticipated dates barring any delays						



References

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UC Davis Children's Hospital

Jorge E. Suarez-Cavelier, MD, FACS is Recognized by Continental Who's Who

PRNewswire/ -- Jorge E. Suarez-Cavelier, MD, FACS, is being recognized by Continental Who's Who as a Top Surgeon for his remarkable contributions in the field of Medicine and acknowledgment of his dedication and commitment as a Cardiothoracic Surgeon with the Advent Health Cardiovascular Institute.

Proudly serving Orlando, Advent Health is committed to delivering quality care to patients from everyday wellness and preventive health care, to life-saving diagnostic services and innovative medical treatments in cancer, and heart failure through the Advent Health Cardiovascular Institute. Patients have direct access to some of the most advanced cardiovascular treatment options available at one of America's most experienced cardiac hospitals.

Board-certified Medical Director and Cardiothoracic Surgeon, Dr. Jorge E. Suarez-Cavelier has garnered a laudable reputation for his vast repertoire of expertise and professional experience in structural heart disease and heart transplants. Fluent in English and Spanish, he offers services in those languages. He has served as Chief of the Department of Cardiothoracic Surgery for over 10 years performing cardiothoracic and vascular surgeries and providing the highest standard of patient-centered care. More specifically, he diagnoses and treats a collection of heart diseases that includes heart failure, coronary artery disease, hypertrophic cardiomyopathy, and congenital heart disease that is obtained through wear and tear, or heart disease that people are born with.

An academic scholar, Dr. Suarez-Cavelier received his Medical Degree and completed his general surgical residency from Javeriana University in Bogota, Columbia. He relocated to the United States for a general surgical residency at Maimonides Medical Center in New York. In light of his academic success he was invited for a Fellowship in vascular and cardiac surgery at Caroline Medical Center, Charlotte, NC, and later invited for a Fellowship in cardiothoracic surgery with Methodist Hospital, Baylor University, Houston, Texas. He has also completed a Fellowship in cardiac surgery under Dr. Albert Starr in Portland, Oregon.

Remaining abreast of the latest developments in his field, he remains a Fellow of the American College of Surgeons and is a member with the Cardiac Surgery Association, Society of Thoracic Surgeons and the Southern Society of Thoracic Surgeons.

Dr. Jorge E. Suarez-Cavelier dedicates his success to his mentors Michael DeBakey, MD, and Francis Robichek, MD.





Offering Hope to Families of Infants with Congenital Differences



Nicklaus Children's Hospital's Fetal Care Center is dedicated to families expecting an infant who will need medical attention immediately after birth. The program offers comprehensive, coordinated care from prenatal diagnosis to delivery, postnatal care and the transition to infant care.

Nurse navigators are available to optimize care access and serve as points of contact for the expectant family, community physicians, pediatricians, perinatologists and neonatologists, as well as coordinate the subspecialty consultations.

Nicklaus Children's offers an array of diagnostic services, including fetal ultrasound, fetal MRI and fetal echocardiography, with the goal to support families in obtaining a definitive diagnosis and plan of care during a single visit. Nicklaus Children's Hospital's subspecialty programs are routinely ranked by *US News & World Report* in its annual listing of "Best Children's Hospitals" in the nation.

We have implemented additional safety precautions, such as arrival screenings and physical distancing measures. For parents who wish to make arrangements for their child to see his or her specialty physician virtually, telehealth options may also be available. For more information visit nicklauschildrens.org/covid19safety



The Fetal Care Center's special delivery unit is a birth center for healthy mothers of high-risk infants who will require immediate access to our team of pediatric intensive and critical care specialists after birth.

- 2 High-technology operating rooms
- 5 Antepartum rooms
- 5 Labor and delivery rooms



**Nicklaus
Children's
Hospital**

Fetal Care Center

To reach a Fetal Care Center nurse navigator, available 24 hours a day, call 1-844-553-3825 | Nicklauschildrens.org/FetalCare

Nicklaus Children's Hospital. For Health. For Life.



Driscoll
Children's Hospital

NEW Opportunity - Pediatric Cardiac Intensive Care

The Driscoll Health System is recruiting a Board Certified/Board Eligible Pediatric Cardiac Intensive Care Physician. Currently, the group consists of six Board Certified Pediatric Critical Care Physicians. The ideal candidate, MD/DO, will be dual boarded by the American Board of Pediatric Critical Care and Pediatric Cardiology or completed additional fellowship training in Pediatric Cardiac Critical Care. Candidates must have extensive expertise in post-operative management of complex and congenital heart patients. The pediatric critical care unit provides a comprehensive range of services, including ECMO, high frequency ventilation, and CRRT.

Successful applicants will enjoy a highly competitive compensation package, medical, dental, and vision, disability and life insurance, excellent retirement plans, generous paid vacation days, paid holidays, paid CME and malpractice insurance.

About Driscoll Children's Hospital

Driscoll Children's Hospital is a 189-bed pediatric tertiary care center with more than 30 medical and surgical specialties offering care throughout South Texas, including Corpus Christi, the Rio Grande Valley, Victoria, and Laredo. Through the vision and generosity of its founder, Clara Driscoll, Driscoll Children's Hospital opened in 1953, becoming the first, and remains the only, free-standing children's hospital in South Texas. In 2019, Driscoll had over 141,000 patient visits, including over 46,000 patients seen at South Texas' first emergency room created exclusively for children.

About Corpus Christi, Texas

Corpus Christi is an excellent place to work, live, play and invest. Enjoy living in a dynamic coastal city with miles of beautiful beaches, world-class hunting, fishing, sailing, and windsurfing. Our mild climate allows for year-round outdoor family activities such as golf, cycling, and tennis. The cost of living is very low, and there is no state income tax!

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Two-Year-Old Palestinian "Miracle Boy" Returns Home After More Than Two Months in Critical Condition in Israel

The Israeli medical team of Save a Child's Heart fought for Hamza's life, treating him and functioning as his parents, while his parents had to stay at home in Ramallah, Palestine due to COVID-19 lockdown

PRNewswire - Hamza was born with life-threatening Congenital Heart Disease and had his first surgery as an infant. The Save a Child's Heart (SACH) medical team decided a few months ago that he needed a second surgery to survive.

On February 24, 2020, Hamza underwent open heart surgery at Wolfson Medical Center (WMC) in Holon, Israel, by the SACH volunteer medical team.

His recovery from this surgery was very complicated. "We tried to extubate Hamza a few times after his surgery, but he had a hard time breathing by himself. We had to connect him to an ECMO machine, and it kept him alive for a few weeks," said Dr. Racheli Sion Sarid, a senior physician at the Pediatric Intensive Care Unit at Wolfson.

In addition to the huge challenge of keeping Hamza alive through this period, the medical team had to face another major difficulty: Hamza's parents, who went home to the West Bank city of Ramallah for a short visit to see Hamza's siblings, could not return to Israel due to the COVID-19 lockdown.

"The whole medical team in the Pediatric Cardiology unit became his parents," said Dr. Ahmed Amer, a Pediatric Resident at WMC. Dr. Amer, an Israeli Arab physician who is part of the SACH medical team, became the main contact person for Hamza's parents since he speaks Arabic. "We all became his family. The nurses did shifts to hold him in their arms and play with him. We did not keep him alone for a minute. A child his age and in his condition needs to be hugged and loved in order to recover and get stronger, and that's exactly what we did."

Dr. Amer spoke with Hamza's parents every day, sent them pictures and videos and updated them on his recovery.

As Hamza's condition improved, he was able to communicate with his parents through video calls. "The first few times he saw them on video he began crying and it took us a long time to calm him down," said Dr. Amer, "But he got used to it and we even celebrated his birthday a couple of weeks ago together with his parents on video."

Hamza is now walking, laughing, and playing, and soon he will be ready to return home to his family in Ramallah. The medical team at the hospital are preparing to say goodbye, knowing they will never forget him and his story.

"His story is amazing," says Dr. Sarid. "It is a story about a medical team who fought tirelessly for the life of a critically ill child, a story of a brave little boy who survived against all odds, and a story of people, of human beings, helping each other regardless of their origin and religion. This is the story of Save a Child's Heart."



Palestinian child, Hamza, and one of the nurses taking care of him.

About Save a Child's Heart

SACH is an Israeli-based international non-profit organization that has provided care to more than 5,400 children from 62 countries in Africa, the Middle East, Asia, Europe, and South America and trained more than 120 medical professionals from these countries.

SACH is committed to the health and welfare of all children, regardless of nationality, religion, color, gender, or financial situation.



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**Division of Pediatric Cardiology
Saint Louis University
School of Medicine
SSM Health Cardinal Glennon
Children's Hospital**

Non-Invasive Cardiologist

We are seeking an additional non-invasive cardiologist to assist our current faculty with the growing number of echocardiographic procedures performed at our institution. Experience in performing and interpreting both transthoracic and transesophageal echocardiograms is required. Participation in outpatient clinic and inpatient care is also required. An interest in clinical research is encouraged. Academic rank will be commensurate with qualifications and experience.

The cardiology division is housed within the Dorothy and Larry Dallas Heart Center at SSM Health Cardinal Glennon Children's Hospital. The Heart Center opened in 2009 and underwent significant expansion in 2016. An active congenital heart surgery program exists, and the hospital houses state-of-the-art operating rooms, neonatal intensive care unit, pediatric intensive care unit, electrophysiology lab, and a hybrid cardiac catheterization lab/operating suite. SSM Cardinal Glennon Children's Hospital is a free-standing children's hospital and is staffed by faculty members of Saint Louis University School of Medicine.

Interested candidates must submit a cover letter, application, and current CV to:

<https://www.slu.edu/working-at-slu.php>

Other correspondence regarding these positions can be sent to:

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Saint Louis University is a Catholic, Jesuit institution dedicated to student learning, research, health care, and service. Saint Louis University is an Affirmative Action, Equal Opportunity Employer, and encourages nominations of and applications from women and minorities.



Pediatric Cardiologist

The Department of Pediatrics at Southern Illinois University School of Medicine is recruiting an M.D. for a fourth pediatric cardiologist at the Assistant or Associate Professor level. Faculty will join a rapidly expanding cardiology program at our Children's Hospital, an 80-bed CHA affiliated pediatric referral center for Central and Southern Illinois with a referral base of almost 2 million. The current program includes state-of-the-art noninvasive imaging in TTE, TEE, fetal echocardiogram, and advanced MRI imaging. We have developed a highly successful collaborative clinical and research program with a nationally recognized pediatric cardiology center. Opportunities exist to participate in resident and medical student education and receive an advanced degree in medical education. Candidates must be board eligible in Pediatrics and Pediatric Cardiology. Illinois licensure is required prior to official start date. Travel in central Illinois to outreach clinics is required.

Applications are accepted online at:

<https://siumed.hiretouch.com/>.

For additional information, please contact:

Ramzi Nicolas, MD

P. 217.545.9706

E. rnicholas@siumed.edu

SIU is an EO/AA employer.



2020

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05

Pediatric Cardiac Fitness and Rehabilitation Programs for Congenital Heart Disease: A Practical Guide

Virtual

<https://bostonchildrens.cloud-cme.com/course/courseoverview?P=3000&EID=1811>

09-12

AIMed 20

Laguna Niguel, CA, USA

<https://ai-med.io/all-events/global-summits/aimed-20/>

11-12

CSI Focus D-HF 2020

Frankfurt, Germany

<https://www.csi-congress.org/dhf>

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11-14

CPP 2021

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