Unveiling the gothic aortic arch and cardiac mechanics: insights from young patients after arterial switch operation for d-transposition of the great arteries

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Abstract

The arterial switch operation (ASO) has become the standard surgical treatment for patients with d-transposition of the great arteries. While ASO has significantly improved survival rates, a subset of patients develop a unique anatomical anomaly known as the gothic aortic arch (GAA). Understanding cardiac mechanics in this population is crucial, as altered mechanics can have profound consequences for cardiac function and exercise capacity. The GAA has been associated with changes in ventricular function, hemodynamics, and exercise capacity. Studies have shown a correlation between the GAA and decreased ascending aorta distensibility, loss of systolic wave amplitude across the aortic arch, and adverse cardiovascular outcomes. Various imaging techniques, including echocardiography, cardiac magnetic resonance imaging, and cardiac computed tomography, play a crucial role in assessing cardiac mechanics and evaluating the GAA anomaly. Despite significant advancements, gaps in knowledge regarding the prognostic implications and underlying mechanisms of the GAA anomaly remain. This review aims to explore the implications of the GAA anomaly on cardiac mechanics and its impact on clinical outcomes in young patients after ASO. Advancements in imaging techniques, such as computational modeling, offer promising avenues to enhance our understanding of cardiac mechanics and improve clinical management.

Introduction

D-transposition of the great arteries (d-TGA) is a congenital heart defect characterized by a discordant connection of the aorta and pulmonary artery [1]. This condition leads to two separate circulatory pathways: deoxygenated blood from the systemic circulation flows into the right atrium, then proceeds to the right ventricle and aorta, ultimately returning to the systemic arterial circulation without receiving oxygen. Meanwhile, oxygen-rich blood from the pulmonary veins returns to the left atrium and then flows to the left ventricle (LV), pulmonary artery, and pulmonary arterial circulation. The arterial switch operation (ASO) has become the standard surgical treatment for d-TGA, involving the anatomical correction of the great arteries [1,2].

Although ASO has significantly improved outcomes for patients

with d-TGA, meticulous follow-up is mandatory to prevent potential long-term complications, such as pulmonary artery stenosis, aortic valve dysfunction, and coronary stenosis. In addition, there are emerging concerns related to the gothic aortic arch (GGA) anomaly in this population. Indeed, one area of interest in the post-operative management of young patients after ASO is the impact of GAA on cardiac mechanics [3]. The GGA is characterized by a high, narrow, and elongated aortic arch, which may have implications for cardiac mechanics and hemodynamics. It can be found after coarctation repair in patients with bicuspid aortic valve, or in patients with genetic syndromes such as Loeys-Dietz syndrome [4,5]. This unique anatomical variant has also been observed in a substantial proportion of patients after ASO and is considered a potential risk factor for adverse outcomes [6].

Studying the GAA and cardiac mechanics in young patients who have undergone ASO is significant for several reasons. First, understanding the prevalence and characteristics of the GAA can help identify patients at risk for developing complications or long-term cardiovascular issues [3]. Second, assessing cardiac mechanics provides insights into ventricular function, myocardial strain, and diastolic function, which are crucial aspects of cardiac health and overall patient outcomes [3,6].

By examining the GGA and cardiac mechanics in this population, clinicians can gain a deeper understanding of the post-operative effects of ASO on cardiac structure and function. This knowledge can guide clinical decision-making, optimize patient management strategies, and improve long-term outcomes for young patients with d-TGA who have undergone ASO.

This review aims to provide insights into the GAA anomaly and its impact on cardiac function in young patients with d-TGA after ASO. By examining the available evidence and exploring potential mechanisms, this review aims to shed light on the significance of cardiac adaptation in GAA to optimize the management of these patients.

Anatomy and physiology of the aortic arch

The aortic arch (AA) is a vital component of the cardiovascular system, responsible for distributing oxygenated blood from the heart to the systemic circulation. It is a curved segment of the aorta that extends from the ascending aorta to the descending aorta, forming an arch-like structure. The normal anatomy of the AA includes three main branches: i) brachiocephalic trunk (innominate artery supplying blood to the right subclavian artery, right common carotid artery, and right vertebral artery; ii) left common carotid artery, which provides blood supply to the left side of the head and neck; iii) left subclavian artery, supplying blood to the left upper limb and part of the brain [1].

The concept of the GGA anomaly refers to a specific anatomical variant characterized by a high, narrow, and elongated AA, resembling gothic architecture, and it is commonly observed in patients who have undergone the ASO [6].

The incidence of GAA in patients after ASO varies across studies. It has been reported to occur in approximately 10% to 20% of patients who have undergone ASO for d-TGA [7,8]. However, the exact prevalence may depend on the specific patient population, the imaging modality used for assessment, and the criteria for defining a GAA.

The etiology and mechanisms contributing to the development of a GAA in patients after ASO are not entirely understood. Several factors have been proposed to play a role in its pathogenesis. One possible explanation is the altered geometry and tension of the aorta following ASO [7]. The repositioning of the aorta during the surgical procedure may result in a different distribution of forces, leading to abnormal arch morphology [9]. Another proposed mechanism is related to the presence of residual intramural tissue within the aortic arch. During the ASO, some remnants of the original AA may be left behind, contributing to the abnormal angulation and shape of the arch [10]. Additionally, genetic and developmental factors may contribute to the formation of a GAA. The embryological development of AA involves complex interactions between various signaling pathways and genetic factors. Disruptions or abnormalities in these processes could potentially lead to the development of an atypical arch shape [10,11].

The GAA anomaly can have a significant impact on cardiac mechanics and hemodynamics. The elongated and narrow shape of the AA may result in altered blood flow patterns and increased vascular resistance. This anomaly can lead to increased afterload on the LV, potential flow disturbances, and abnormal distribution of blood to the systemic circulation. These hemodynamic changes may affect cardiac function, exercise capacity, and overall cardiovascular performance [1,2,12].

Cardiac mechanics in the aortic arch anomalies

The presence of a GAA anomaly can influence several aspects of cardiac function [2]. Hemodynamically, the altered geometry can affect blood flow dynamics and create potential turbulence or disturbances in aortic flow patterns. These hemodynamic changes can impact LV afterload, blood pressure, and the overall efficiency of cardiac output [13,14]. Nevertheless, the altered flow patterns in the GAA can lead to variations in shear stress distribution along the aortic wall. Increased shear stress in certain regions of the aorta can trigger pathological remodeling processes, including arterial stiffness, endothelial dysfunction, and vascular remodeling. These changes may further impact LV workload, ventricular-vascular coupling, and cardiac performance [15,16]. Moreover, the GAA anomaly can influence ventricular-vascular interactions, particularly concerning ventricular load and contractility. The altered afterload and flow dynamics can affect left ventricular systolic function, myocardial energetics, and ventricular-arterial coupling. These interactions may have implications for ventricular efficiency, myocardial oxygen demand, and overall cardiac performance [17,18].

Therefore, while ASO successfully corrects the anatomical abnormalities associated with d-TGA, it can introduce changes in cardiac mechanics that may have implications for long-term cardiovascular function in these patients [3,6]. Follow-up of patients after ASO includes serial echocardiographic studies to evaluate LV and AA remodeling. This remodeling process can lead to changes in ventricular geometry, wall thickness, and chamber size, potentially impacting cardiac mechanics [19,20].

While some surgical strategies may help prevent the development of GAA [21], no formal recommendations exist about the optimal treatment (surgical or percutaneous) in cases of significant functional obstruction [22], although a case of successful stenting implantation has been described and treatment of systemic hypertension is mandatory [23].

Imaging modalities for assessing cardiac mechanics

Transthoracic and transesophageal echocardiography play a crucial role in assessing cardiac adaptation in this patient population. These techniques allow for the evaluation of ventricular dimensions

and function. Additionally, advanced echocardiographic methods such as tissue Doppler imaging and strain imaging provide quantitative assessments of ventricular function, including strain, strain rate, and myocardial deformation indices [12].

The presence of reduced LV function after ASO has been demonstrated by several studies. Some have shown that young patients after ASO can exhibit preserved LV systolic function with only subtle abnormalities in myocardial mechanics [12,24-26]. Other studies showed that alterations in ventricular function can occur after ASO, particularly in the right ventricle (RV) [19]. Additionally, changes in ventricular geometry, such as increased ventricular mass and altered ventricular shape, have been observed [27].

Myocardial strain has emerged as an important parameter for assessing cardiac mechanics in pediatric patients with different cardiac diseases [28,29]. It represents the deformation or change in myocardial tissue during the cardiac cycle and provides insights into myocardial contractility and function [30]. Studies investigating myocardial strain in patients with GAA after ASO demonstrated alterations in strain values and distribution patterns, especially in LV basal segments [26]. This could be due to higher wall stress in these segments, according to Laplace's law. Some studies have reported decreased strain values, indicating impaired myocardial contractility, also in the RV [31,32]. Changes in strain distribution, including regional variations in strain values, have also been observed, suggesting regional differences in myocardial function [33,34]. Figure 1 shows strain abnormalities and GAA as assessed by transthoracic echocardiography in a patient with d-TGA, years after the ASO operation.

Diastolic dysfunction can be observed in patients with d-TGA after ASO [24]. The altered geometry of the LV and AA may impact diastolic filling dynamics. Diastolic dysfunction can manifest as impaired relaxation, increased stiffness, or abnormal filling patterns. Evaluation of diastolic function using echocardiography or other imaging modalities can provide valuable insights into cardiac mechanics and potential consequences for patients' clinical outcomes. Studies showed that diastolic dysfunction can occur particularly in the early postoperative period [24]. Changes in diastolic parameters, such as decreased early diastolic velocities and increased myocardial stiffness, have also been observed [2,35].

Other imaging modalities, such as cardiac magnetic resonance

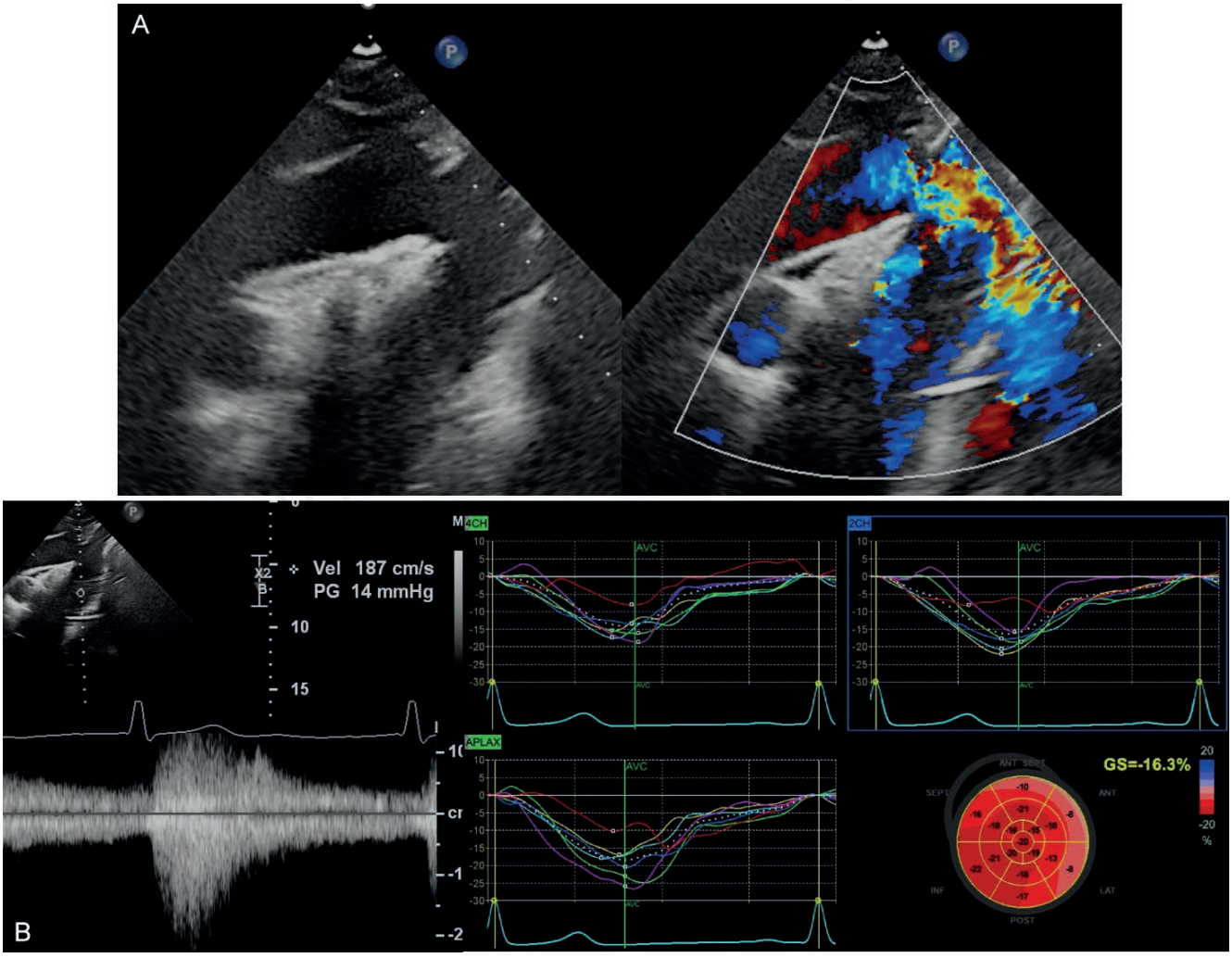

Figure 1. A) Gothic aortic arch as visualized by 2D echocardiography, with and without color Doppler; B) trans-isthmic gradient as assessed by continuous-wave Doppler (left); left ventricle global longitudinal strain, showing mild impairment of longitudinal function, especially involving basal segments.

(CMR) and cardiac computed tomography (CCT), may add valuable information in evaluating patients with GAA (Figure 2). CMR offers excellent spatial resolution and tissue characterization, making it particularly valuable for assessing the complex anatomy of the aorta, pulmonary arteries, and coronary arteries after ASO [35]. It allows for accurate visualization of the neoaortic root and the presence of any stenosis or abnormal anatomy [36,37]. Furthermore, CMR allows for the assessment of blood flow patterns, particularly in anomalous vessel anatomy, providing crucial information for planning further interventions or surgical procedures [38,39]. CMR has also proven to be a valuable tool in the long-term follow-up of patients after ASO, allowing for the detection of potential complications such as neoaortic root dilation or stenosis. Additionally, serial CMR examinations can provide valuable information on the progression of ventricular remodeling or the development of any late complications [40]. Lastly, with the development of 4D flow CMR, various parameters such as helicity, turbulent kinetic energy, and wall shear stress have been introduced to investigate flow patterns in different cardiac diseases, including aortic aneurysm and coarctation [41,42]. This technique might also help patients with GAA to better understand blood flow patterns, detect subtle abnormalities in aortic blood flow even before structural anomalies become apparent, and stratify patients based on their risk of aortic complications. Also, by combining 4D flow MRI with computational modeling and AI, clinicians can create patient-specific simulations of blood flow and vortex behavior in the aortic arch. This personalized approach can aid in tailoring treatment plans and optimizing outcomes for each patient [43].

CCT is employed for anatomical assessment and evaluation of coronary artery anatomy in patients with d-TGA after ASO [44]. It allows for accurate visualization of the aorta, pulmonary arteries, and great vessels. However, it involves radiation exposure and is less commonly utilized for assessing cardiac mechanics compared to echocardiography and CMR.

Table 1 discusses the advantages and limitations of each modality, including echocardiography, CMR, and CCT. Indeed, by integrating echocardiography, CMR, and cardiac CT, clinicians can obtain a more comprehensive evaluation of GAA abnormalities. Each imaging modality contributes unique information, allowing for a better understanding of the complex anatomy, hemodynamics, and functional consequences of these anomalies [45].

Clinical implications and long-term outcomes

The presence of the GAA anomaly has clinical implications for cardiac mechanics and hemodynamics [7,46]. These alterations in cardiac mechanics may impact exercise capacity, with potential limitations in exercise tolerance and performance [47,48]. Long-term outcomes in patients with GAA are not yet fully understood, but there is evidence suggesting an association with an increased risk of adverse cardiovascular events and the need for re-interventions [36,47]. Bruse *et al*. found that patients with a more normal-shaped AA had significantly better LV-indexed end-diastolic volume, LV-indexed mass, and LV function compared to those with abnormal AA shapes after 12-38 years from operation of coarctation of the aorta (CoA) [49]. In the long-term follow-up after repair of CoA, it has been observed that a GAA shape is associated with reduced distensibility of the ascending aorta and increased loss of systolic wave amplitude across the AA [50]. Managing these patients poses challenges, including the need for comprehensive and specialized follow-up care, monitoring of cardiac function, and timely interventions [7,47]. Potential strategies for optimizing outcomes in these patients include close monitoring of cardiac function through multimodality imaging, early identification of complications, and timely intervention when indicated [7,48].

Future directions and research perspectives

Despite advances in understanding the GAA anomaly and its impact on cardiac anatomy and function, there are still several gaps in our knowledge. First, the long-term implications of altered cardiac mechanics and the specific factors influencing clinical outcomes in this patient population require further investigation [49]. Longitudinal

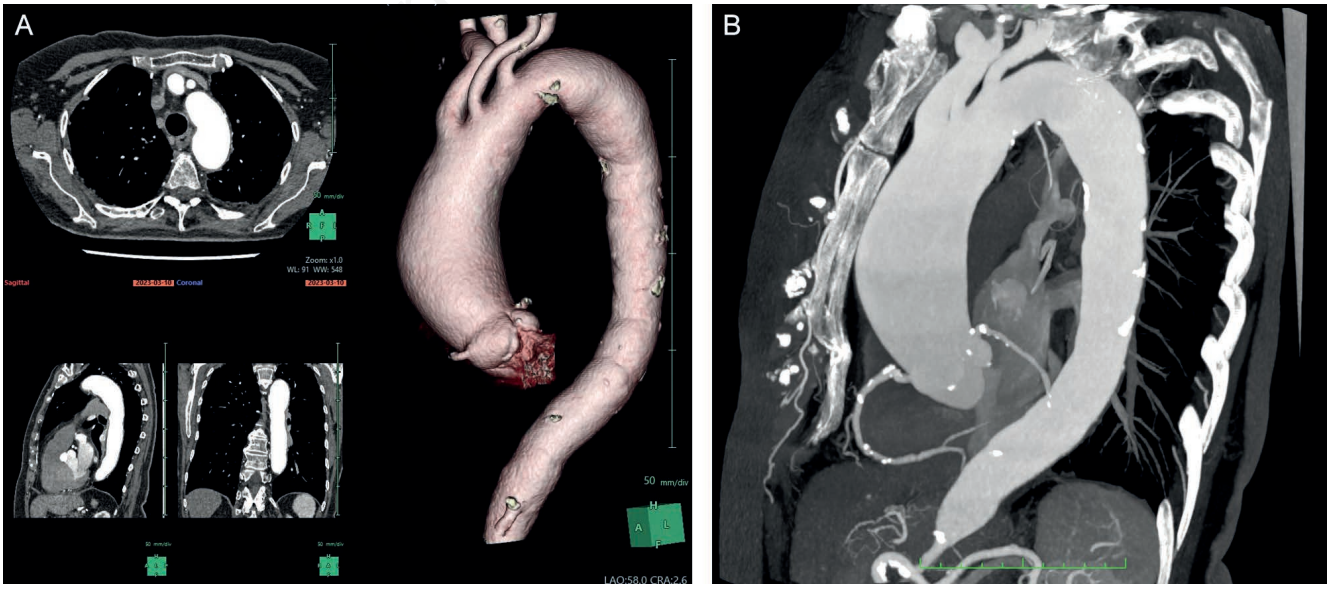

Figure 2. A) Volume rendering technique; B) multiplanar computed tomography reconstruction showing gothic aortic arch morphology.

Table 1. Advantages and limitations of echocardiography, cardiac magnetic resonance imaging, and cardiac computed tomography.

Imaging modality	Advantages	Limitations
Echocardiography	1. Widely available, portable, and cost-effective. 2. Real-time imaging, dynamic assessment of cardiac function and hemodynamics. 3. Excellent temporal resolution, enabling the assessment of valvular function, wall motion abnormalities, and blood flow patterns. 4. Multiple imaging planes, facilitating comprehensive evaluation of cardiac structures. 5. Assess myocardial strain and deformation using advanced imaging techniques such as speckle-tracking echocardiography.	1. Limited acoustic window in some patients, hindering visualization of specific cardiac structures. 2. Operator-dependent, requiring expertise for accurate interpretation and measurements. 3. Relatively lower spatial resolution compared to cardiac MRI and CT. 4. Limited assessment of extracardiac structures and calcifications.
Cardiac magnetic resonance imaging	1. Excellent soft tissue contrast and detailed anatomical visualization. 2. Comprehensive assessment of cardiac function. 3. Precise evaluation of myocardial tissue characteristics. 4. Safe for repeat examinations, especially in young patients. 5. Enables the assessment of blood flow patterns and vascular anatomy.	1. Longer examination time compared to other modalities. 2. Limited its use in patients who cannot tolerate prolonged scanning. 3. Contraindicated in patients with certain metallic implants or devices. 4. Limited availability and higher cost compared to echocardiography and cardiac CT. 5. Limited assessment of coronary artery.
Cardiac computed tomography	1. Excellent spatial resolution and fast acquisition times, allowing for detailed anatomical evaluation. 2. Accurate assessment of coronary artery anatomy. 3. 3D reconstructions, facilitating visualization of complex cardiac structures and providing additional information for surgical planning. 4. Provides detailed assessment of extracardiac structures, including great vessels and thoracic anatomy.	1. Exposure to ionizing radiation, limiting its use in young patients, and requiring consideration of radiation dose reduction strategies. 2. Requires iodinated contrast media administration, which may be contraindicated in patients with renal dysfunction or iodine allergy. 3. Limited assessment of myocardial tissue characteristics compared to cardiac MRI. 4. Lower temporal resolution compared to echocardiography and cardiac MRI. 5. Limited evaluation of valvular function and flow dynamics compared to echocardiography and cardiac MRI.

CT, computed tomography; MRI, magnetic resonance imaging.

studies with larger sample sizes and longer follow-up periods are needed to assess the progression of cardiac dysfunction and its impact on morbidity and mortality. Additionally, there is a need for comparative studies that explore the differences in cardiac mechanics and outcomes between patients with different types of AA anomalies.

Potential for advanced imaging techniques and computational modeling

Advanced imaging techniques, such as 3D echocardiography, vortex analysis with different imaging modalities, CMR, and CCT, have shown promise in providing detailed information on cardiac mechanics in patients with GAA abnormalities. Integration of these imaging modalities with computational modeling approaches, such as finite element analysis and fluid dynamics simulations, can further enhance our understanding of the AA and its surrounding structures and the complex interactions between the altered cardiac mechanics, blood flow patterns, and clinical outcomes [50-52]. Also, patient-specific 3D models can be created, and surgeons can use these models to plan surgeries, simulate different treatment scenarios, and optimize their strategies for each patient, leading to more precise and personalized care. This can not only reduce the time required for diagnosis but could also enhance the reliability of results. Machine learning models may also predict the likelihood of complications and guide clinicians and surgeons in making informed decisions regarding treatment options and timing.

To improve outcomes in young patients with GAA after ASO, there is a need for targeted clinical studies and therapeutic interventions. Prospective studies should focus on identifying early markers of cardiac dysfunction and developing risk stratification models to guide clinical management. This may include tailored approaches, such as personalized AA reconstruction techniques or stenting, to improve blood flow dynamics and reduce cardiac workload [50,51]. Collaboration between multidisciplinary teams comprising cardiologists, cardiac surgeons, imaging specialists, and computational modelers is crucial for developing comprehensive treatment strategies and optimizing outcomes [53,54].

Conclusions

This review highlights the significant role of cardiac mechanics in young patients after ASO for d-TGA. The presence of a GAA anomaly in these patients has been associated with altered cardiac mechanics, including changes in ventricular function and hemodynamics, which may have clinical implications, such as reduced exercise capacity and an increased risk of adverse cardiovascular outcomes. Understanding and accurately assessing cardiac mechanics is crucial for the optimal clinical management of these patients. Multimodality imaging techniques, including echocardiography, CMR, and CCT, play a vital role in providing comprehensive assessments. However, there are still gaps in our knowledge, and further research is needed to advance our

understanding, refine diagnostic techniques, and develop targeted therapeutic interventions. This will ultimately lead to improved outcomes and a better long-term prognosis for young patients with GAA abnormalities after ASO for d-TGA.

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