The utility of speckle-tracking echocardiography in early and midterm follow-up after anomalous origin of the left coronary artery from the pulmonary artery repair

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ABSTRACT

Anomalous left coronary artery from the pulmonary artery (ALCAPA) is a reversible cause of left ventricular (LV) dysfunction in infants. The LV function is expected to improve serially and return to normal by 1 year after surgical repair. The pattern of improvement in LV function has not been serially analyzed after ALCAPA repair. We report our preliminary experience with serial assessment of LV function in infants undergoing ALCAPA repair utilizing speckle tracking echocardiography.

Keywords: Anomalous origin of the left coronary artery from the pulmonary artery, functional echocardiography, global longitudinal strain

INTRODUCTION

Anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA) is a congenital heart disease with an estimated incidence of 1 in 300,000.[1] The left coronary artery arises from the pulmonary artery which supplies the low-pressure pulmonary vascular bed. This causes reversal of flow from the anomalous left coronary artery to the pulmonary trunk and a "coronary steal" phenomenon resulting in myocardial insufficiency.[2] Infants with ALCAPA often have severe left ventricular (LV) systolic dysfunction. The LV systolic function improves after surgical repair in a vast majority of children.[3] Published data suggest that the LV function returns to normal by 1 year after the surgical repair.[4] It is believed that chronic insufficiency results in a myocardium that is hypocontractile but whose viability is preserved – a so-called "hibernating myocardium" in those children whose LV function recovers after surgery.[5] Speckle-tracking echocardiography (STE) has the potential to predict improvement or deterioration of LV function earlier than global markers such as ejection fraction (EF).[6] Most published literature has reported LV function assessment at a single postoperative visit.[7] There are no published data on the serial assessment of LV function after ALCAPA repair. We reviewed our early experience with serial monitoring of LV function after ALCAPA repair in infants by comprehensive echocardiographic evaluation.

METHODS

This was a retrospective case record analysis of all patients who were diagnosed in our center to have ALCAPA between March 2020 and September 2022.

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All echocardiograms were performed by the same pediatric cardiologist (MRK). The echocardiograms were electrocardiography gated and performed on an Epiq Elite ultrasound machine (Philips Medical Systems, Andover, MA, USA) using a S9-2 single-crystal transducer. STE was performed online using the AutoStrain LV package (TomTec Systems, Munich, Germany). All patients underwent coronary translocation through a median sternotomy.

RESULTS

During the study period, eight infants were diagnosed to have ALCAPA at our center. The family of three infants opted against surgical repair. Four of the five operated infants were female, and the median age was 3 months (50 days to 5 months). The median duration of postoperative mechanical ventilation was 4 days (3–8 days), and the median intensive care unit stay was 6 days (5–13 days). All infants were discharged home on diuretics, aldosterone antagonists, angiotensin-converting enzyme inhibitors, and heart rate-lowering medications. At 1 year follow-up, enalapril was continued in patient 4, whereas all medications were discontinued in the other children.

The preoperative as well as the postoperative LV function assessment is tabulated in Table 1. The immediate postoperative and 1-month postoperative assessment was not possible for patient 1 because of the strict travel restrictions related to the first lockdown during the pandemic.

The median EF and global longitudinal strain (GLS) at the various assessment periods are depicted in Figure 1a and b. The values at each visit were compared to the preoperative values by Wilcoxon signed-rank test. Statistical significance was reached by 6 months ($P = 0.043$ for GLS compared to baseline and 0.043 for EF compared to baseline) and was sustained at 1 year ($P = 0.038$ for GLS for 0.033 for EF).

Patient 2 was illustrative of the utility of GLS in follow-up after ALCAPA repair. The GLS was very low, and the bull's eye showed akinesia/severe hypokinesia of the anterior segments [Figure 2a]. In the post-operative echocardiogram, there was a modest improvement in the GLS and a small but apparent improvement in the segmental scores of the affected segment [Figure 2b]. The subsequent analysis showed progressive improvement in regional contractility, with all segments exhibiting normal contractility by 1 year [Figure 2c-e and Videos 1-5].

Patient 4 had a very slow improvement in the EF during the early postoperative period. However, there was a consistent improvement in the GLS, which was reassuring.

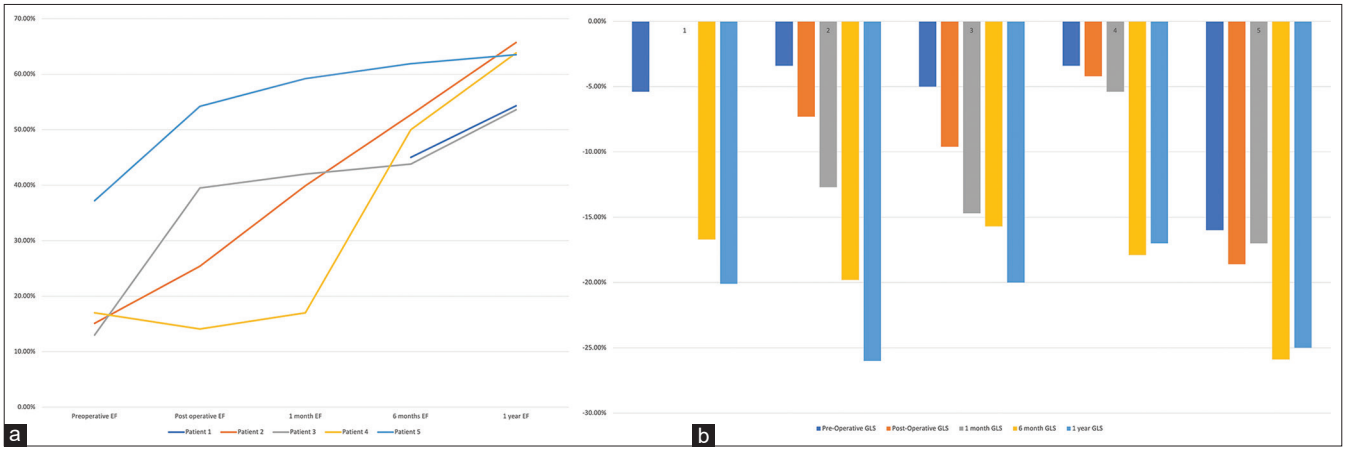

Figure 1: (a) Line plot depicting the serial improvement in ejection fraction in each infant after surgical repair. (b) Clustered column chart depicting the serial improvement in global longitudinal strain (GLS) in each infant

Table 1: Ejection fraction and global longitudinal strain values of each of the patients during the preoperative echocardiogram, postoperative echocardiogram, and follow-up echocardiograms 1, 6, and 12 months after surgery

Patient number	Preoperative EF (%)	Preoperative GLS (%)	Postoperative EF (%)	Postoperative GLS (%)	1-month EF (%)	1-month GLS (%)	6-month EF (%)	6-month GLS (%)	1-year EF (%)	1-year GLS (%)
1	9.60	−5.40					45	−16.70	54.30	−20.10
2	15.10	−3.40	25.40	−7.30	39.90	−12.70	52.70	−19.80	65.7	−26
3	13	−5	39.50	−9.60	42	−14.70	43.80	−15.70	53.60	−20
4	17	−3.40	14.10	−4.20	17	−5.40	50	−17.90	63.8	−17
5	37.20	−16	54.20	−18.60	59.20	−17	61.90	−25.90	63.5	−25

EF: Ejection fraction, GLS: Global longitudinal strain

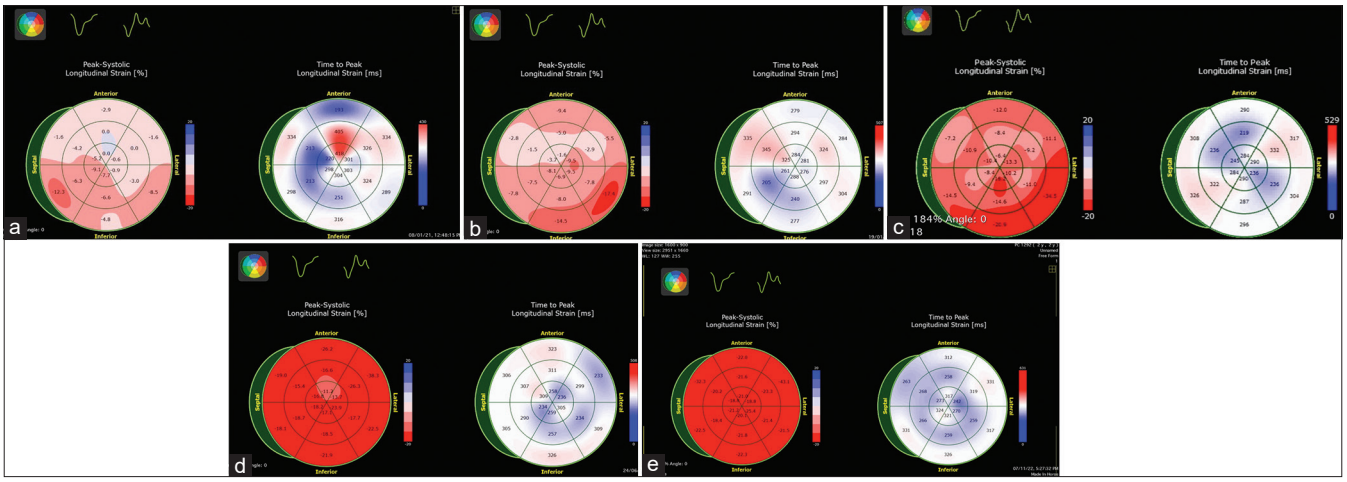

Figure 2: (a-e) Bull's eye of the longitudinal speckle tracking echocardiography of patient 2 on the pre-operative (a), post-operative (b), 1 month (c), 6 month (d) and 1 year (e) demonstrating serial improvement in regional hypokinesia

Patient 5 highlighted one of the drawbacks of GLS, which remains an afterload-dependent modality. This infant had severe mitral insufficiency. Although the infant had symptomatic heart failure and poor LV contractility, the LVEF was 37%. This was likely an overestimation of LV contractility because of the decreased afterload. Correspondingly, the GLS was also near normal at –16%.

DISCUSSION

This is the first study to serially assess global LV function and longitudinal myocardial deformation in infants with ALCAPA before and after surgical repair. Our study shows that infants with ALCAPA demonstrate improvement in LV function and myocardial deformation indices immediately after surgical repair and these indices continue to improve in the 1st year after surgical repair.

In contemporary series, it is evident that repair of ALCAPA can be performed with low mortality and that the LV function recovers in a vast majority of patients.[8] In one of the earliest series of ALCAPA repair, Schwartz *et al.* noted that LV function normalized by 1 year after surgery with a median time to recovery of 2–7 months.[5] In a series of 29 patients reported by Ismail *et al.*, 90% underwent coronary translocation with no mortality at 1-year follow-up. Improvement in LV function was apparent by 6 weeks and normalized by 1 year of age.[4] The largest and most contemporary cohort of ALCAPA was reported by Radman *et al.* There were 170 patients who survived to discharge and the LV function normalized in 79% by 1 year and 97% by 3 years.[9] However, these studies used the motion mode (M-mode) echocardiography to assess LV function, and the trajectory of LV function improvement in the 1st year after surgery was not reported.

Cabrera *et al.* analyzed LV function by STE in a cohort of 14 patients who had undergone ALCAPA repair. A single postoperative echocardiogram study was performed in patients 2–16 years after ALCAPA repair. They noted decreased GLS in 50% of children post-ALCAPA repair.[7] Although the results of this study are contradictory to our results, the patients were not followed up serially, and hence, no information is available on the preoperative as well as early postoperative GLS.

Di Salvo *et al.* performed STE on 30 patients who underwent ALCAPA repair and compared the GLS with 16 age-matched controls.[10] They demonstrated that the GLS was significantly lower in ALCAPA patients than controls. Naqvi *et al.* compared the myocardial deformation analysis of 20 children who underwent ALCAPA repair. This showed severe impairment in the preoperative echocardiogram with improvement in the postoperative echocardiogram performed at a median time period of 1.7 years after surgical repair.[11] These myocardial deformation studies were performed retrospectively and only using a single apical four-chamber view instead of the composite three views for longitudinal function assessment. None of the previous studies were performed on the AutoStrain LV strain package. Differences in GLS values between different vendor software have been reported.[12]

We acknowledge some important limitations in our study. The most glaring shortcoming is the very small sample volume. There are no published age-specific normative values for GLS in Indian children, and we, hence, did not comment on the normalization of GLS but instead focused on the improvement of GLS during serial examinations in each child. The person performing STE was not blinded to the clinical details of the patient, and hence, there remains a potential for bias in the measurements. Our data also do not identify preoperative predictors for recovery of LV function, which may be more useful while counseling families.

Despite these limitations, our preliminary experience suggests that STE, in general, and GLS, in particular, could potentially have an important role in follow-up of infants after surgical repair of ALCAPA. A multicenter prospective study involving major pediatric cardiac centers in the country could provide additional data to support our findings.

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Conflicts of interest

There are no conflicts of interest.

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