Cardiology in the Young

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Original Article

Cite this article: Dennis MR, Cusick A, Borilovic J, Nicholson C, Derwin T, Puranik R, and Celermajer DS (2019) Left arm structure and function late after subclavian flap repair of aortic coarctation in childhood. *Cardiology in the Young* **29**: 856–861. doi: 10.1017/S1047951119000386

Received: 26 November 2018
Revised: 24 January 2019
Accepted: 9 February 2019
First published online: 20 June 2019

Key words:
CHD; muscle mass; grip strength; function

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Left arm structure and function late after subclavian flap repair of aortic coarctation in childhood

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Abstract

Objectives: Concerns exist over the long-term consequences of subclavian artery ligation in subclavian flap repair for coarctation of the aorta. We sought to analyse upper limb structural and functional performance in adults who have had surgery in childhood for coarctation of the aorta, using either subclavian flap repair or end to end aortic anastomosis. *Methods:* Two-group observational design using anatomical and upper limb functional performance measures. Purposive sampling from our specialist adult congenital heart disease database of patients who received subclavian flap repair or end to end anastomosis for coarctation of the aorta as children. Upper limb measurements were completed using MRI and blood flow velocity with ultrasound imaging. Bilateral standardised upper limb functional testing of assessment of strength, dexterity and a standardised self-report of upper limb disability was completed. *Results:* Eighteen right-handed patients, 9 with subclavian repair, (38 ± 12 years, 78% males) were studied. Age at repair was 4.7 ± 5.9 years; mean time from initial repair 32 ± 9 years. The subclavian group had a larger difference between right and left when compared the end to end anastomosis group in: lower arm muscle mass (94.5 ± 42.3 mls versus 37.8 ± 94.5 mls, $p = 0.008$), lower arm maximal cross-sectional area, (5.9 ± 2.8 cm^2 versus 2.9 ± 2.6 cm^2, $p = 0.038$) and grip strength (14.7 ± 8.3 lbs versus 5.9 ± 5.3 lbs, $p = 0.016$) There were no significant functional differences between groups. *Conclusions:* In adults with repaired coarctation of the aorta, those with subclavian flap repair had a greater right to left arm muscle mass and grip strength differential when compared to those with end to end anastomosis repair.

Introduction

Coarctation of the aorta is a relatively common congenital defect, affecting 1 in 2500 live births. Corrective surgery was first performed in 1944 and a variety of techniques have been employed to relieve the stenosis, with now excellent post-operative survival.[1–4] The subclavian flap repair, in which the subclavian artery is ligated distally and the proximal end reattached over the coarctation to relieve the obstruction,[5] has been used for decades. Despite this, there have been concerns about the possible long-term consequences of this on the left arm.[2] Reported effects on vascular supply, strength and function have been variable and limited by subjective self-reporting,[1,6] child or relatively short-term follow up.[7–9] To date, research on long-term structural and functional sequelae of subclavian flap repair in an adult population has been scant and limited to non-standardised data collection processes.

We aimed to analyse upper limb structural and functional parameters in adults who had surgery in childhood for coarctation of the aorta using either subclavian flap repair or end to end anastomosis. In this latter group, vascular supply to the left arm is unaffected. We examined between-limb differences, in right-handed patients, comparing the consequences of the two different types of coarctation repair procedure.

Methods

Study population

This single centre prospective study was performed at a quaternary level adult congenital heart disease centre in Sydney, Australia. Through our adult congenital heart disease database, patients >16 years of age with coarctation of the aorta repaired with either subclavian flap repair or end to end anastomosis were identified. Those with concomitant complex CHD were excluded and only right-handed subjects were included. Patients were consecutively contacted in alphabetical order and recruited into the study until a total of 18 patients were consented and enrolled (9 subclavian flap, 9 end to end anastomosis). Patients with re-intervention on the coarctation after initial repair, or significant residual obstruction (defined as blood pressure arm versus leg difference of >20 mm Hg) were excluded. The study was completed under the Sydney Local Health District, Ethics Research and Governance, Protocol Number X17-0012.

Study design

A prospective observational two-group design was used to compare structural (muscle mass, bone length and blood flow) and functional attributes of these adults. All patients underwent MRI, ultrasound imaging of upper extremity blood flow and functional assessments, as outlined below.

Outcomes: We prospectively defined one primary structural and one primary functional endpoint. The primary structural outcome was the difference in forearm muscle mass between right and left arms, comparing differences between the subclavian flap and end to end anastomosis groups. It was hypothesised that this right–left difference would be significantly greater in patients after subclavian repair compared to end to end anastomosis. The primary functional endpoint was the difference in grip strength between right and left arms between groups. Secondary outcomes included differences in bone length, blood flow, grip strength and functional metrics, between the right and left arms comparing the two groups.

Power and recruitment: Previous studies have shown a 4% average difference in muscle thickness associated with hand dominance.[10] Therefore, power calculations were made assuming a 5% difference in forearm muscle thickness (right minus left) in end to end anastomosis and a 20% difference (right minus left) in the subclavian flap group. A 10% SD for right–left difference was assumed in each group. To achieve 80% power at the $2p < 0.05$ significance level, seven subjects per group were required. Allowing for drop-outs, technically inadequate scans and lack of precision around the assumptions above, we aimed to enrol nine subjects per group.

General assessment

Age, height, weight and hand dominance were ascertained. Past medical history was taken assessing for the presence of hypertension, diabetes, smoking history, self-reported substance abuse or any other medical condition that could impact arm functioning.

MRI protocol for bone length and muscle mass assessment

MRI scans were taken using a 1.5 T Phillips imaging unit at specialist MRI (sMRI). The patient lay supine with arms extended at their side, 2×4 Channel Flex Coils (1 Large 1 Medium Size) and the in-built bore body coil were used for the upper arm. An 8 Channel SENSE Body Coil was utilised for the lower arm. Localiser images were obtained. For bone length measurement and anatomical land marking, T1-weighted coronal images (FOV 400, slice thickness 3 mm, 1 mm Gap, TR 430-650, TE 10) of the upper arm and sagittal T1-weighted images of the forearm were completed (FOV 350, slice thickness 3 mm, 1 mm gap, TR 430-650, TE 10). For muscle measurement of the upper arm; cross-sectional T1-weighted images using a spin echo sequence were obtained (FOV 200, slice thickness 10 mm, no gap, TR 400-700, TE 10) from the mid-point of the elbow joint to 15 cm proximally. For the lower arm, cross-sectional images T1-Weighted images (FOV 150, slice thickness 10 mm, no gap, TR 400-700, TE 10) were obtained from the metacarpal phalangeal joints to above the olecranon process. Data was then analysed using Osirix MD v9.5.1, Pixmeo Sarl, Bernex, Switzerland. All MRI analyses were completed by a trained cardiac magnetic resonance imaging cardiologist, blinded to patient, hand dominance, demographics and type of coarctation repair (MD). Inter-rater reliability was assessed by RP completing two studies blinded with a difference <5%.

Bone length measurements: Bone length measurements (humeral, radial and ulnar) were taken using MRI in the equivalent positioning to gross specimen measurements as per established practice – Buikstra and Ubelaker.[11]

Muscle mass measurements: On each slice, muscle tissue boundaries were traced, with bone and fat excluded. In the upper arm, slices were included for muscle mass measurement from 15 cm proximal to the mid-point of the elbow joint to, but not including the first slice where the head of radius was visualised. For the forearm, the first slice including head of the radius to the radiocarpal joint was included for muscle mass measurement. Muscle volume was calculated by the summation of areas of all the slices automatically taking into account the slice thickness. Maximal cross-sectional area (MCSA) was defined as the maximal slice area measured, excluding bone and adipose tissue.

Ultrasound blood flow protocol

Vascular ultrasound was used assess blood flow velocity and endothelial function through the left and right arms using a modified flow mediated dilatation (FMD) protocol as previously described.[12] Patients were asked to refrain from caffeine, 24 hours prior to the procedure. The examination was performed in a temperature-controlled room (20–25°C) with subjects resting in a supine position. Blood pressure measurements were taken a minimum 10 minutes prior to commencement of tests. Vessel flow was assessed using ultrasound measurement of the brachial artery diameter and changes in brachial artery flow (7–12 MHz linear array, Logic 7, GE). In subclavian flap repairs in the left arm, the largest vessel at or proximal to the antecubital fossa was used. A 9 cm pneumatic cuff is placed around the forearm immediately below the elbow and a 4 cm length of the brachial artery was imaged in a longitudinal section above the antecubital fossa. Arterial diameters and flow velocities were recorded before and after cuff inflation/deflation.

Physical capability and functional performance assessment

Functional performance was indicated by using standardised tests of strength, manual dexterity, bimanual coordination and unilateral gross manual dexterity. All testing was conducted by registered occupational therapists (AC & JB) who trained in the protocol to ensure consistent administration and scoring. Inter-rater agreement was assessed across all measures with 5 of the 18 participants having data collected simultaneously by both raters, independently scored and results compared.

Physical capability – strength: Handgrip strength was assessed using the Jamar™ hydraulic hand dynamometer with a standardised protocol as per guidelines[13] including self-selected handgrip, standing position with the same leg-stance, forearm and elbow position across participants and within trials. The same dynamometer was used across all participants. The mean score of three trials was used as the final score for each limb. Inter-rater agreement was 100%.

https://doi.org/10.1017/S1047951119000386 Published online by Cambridge University Press

Fine motor manual dexterity: The Jamar™ Nine Hole Peg Test (NHPT)[14] model of this standardised timed observed test of finger dexterity was used.[15] Participants completed the test three times using alternating limbs. The mean time was used as the final score for each limb. The time was read off the digital stopwatch which showed minutes and seconds to the third decimal place; one decimal place was used. Test time commenced with the verbal instruction to start and finish time was taken when the last peg hit the holding-dish. Inter-rater agreement using full seconds only was 100%.

Unilateral gross-motor dexterity: The standard Box and Blocks test (BBT) is an observed timed performance test that measures unilateral gross manual dexterity[16] and is one of the most commonly used assessments of upper limb function across a wide variety of clinical populations.[17] A commercially available wooden BBT was used (Sammons Preston Model). Participants completed the test three times using alternate limbs. The number of blocks moved correctly in each trial was counted and the mean taken as the final score. Inter-rater agreement was 100%.

Manual dexterity and bi-manual coordination: The Perdue Peg Board Test (PPB)[18] is a standardised observed timed performance test measuring fine and gross movements of the fingers, hands and arms as well as fine fingertip dexterity. A commercially available PPB was used (Lafayette Instrument Model 32020A). Between one and three complete PPB trials were completed by each participant; variation in trial number was a result of testing scheduling. The mean of trials completed was used for final scores.

Activity participation: Activity participation was assessed using the Quick-DASH[19] standardised self-report measure developed to describe disability experienced by respondents and monitor symptoms and function. The freely available Quick-DASH disability/symptoms section (11 items) was obtained and used in accordance with conditions of use.

Statistical analysis

Statistical analysis was completed using SPSS version 25.0 (IBM Inc., Armonk, NY, United States of America). Continuous variables were summarised as mean and standard deviation or median with inter-quartile range. Student t-test was used for comparison of continuous variables. Categorical variables were summarised as frequencies and percentages. Comparison of categorical variable groups was performed using chi-squared test or Fischer exact test, where applicable. A two-tailed value of $p < 0.05$ was considered statistically significant.

Results

All 18 patients (38 ± 12 years, 78% males) were independent in activities of daily living and underwent ultrasound and functional assessment. Seventeen (nine subclavian flap and eight end to end anastomosis) completed MRI assessment (one subject was excluded owing to presence of an implantable cardiac defibrillator). The mean age at coarctation repair was 4.7 ± 5.9 years (range 0.1–18 years); and was similar between groups (subclavian flap 3.9 ± 5.9 years, end to end anastomosis 5.9 ± 6.2 years, $p = 0.62$). The mean time from initial repair was 32 ± 9 years and there was no significant difference in follow-up periods between groups (33 ± 6 years – subclavian flap, 32 ± 12 years – end to end anastomosis; $p = 0.73$). At the time of study, no patient had significant morbidity that would impact study assessment. No patient reported symptoms suggestive of ischaemia on rest or exercise.

Overall cohort

Right arm blood pressure, bone length and muscle mass metrics were all significantly greater than the left arm in all subjects (Supplementary Table 1), except for radial bone length and the percentage change in the FMD.

End-to-end anastomoses versus subclavian flap repair (between-group comparison)

Full Comparison of the end to end anastomosis cohort and subclavian flap cohort is shown in Table 1. There were no significant differences in the bone or muscle metrics between the end to end anastomosis and subclavian flap cohorts. Mean diastolic blood pressure of both arms were significantly higher in the end to end anastomosis group in right (84 ± 9.6 mm Hg versus 75 ± 10 mm Hg; $p = 0.017$) and the left arms (79 ± 12.5 mm Hg versus 66 ± 9.0 mm Hg; $p = 0.018$).

Within-group differences comparison: end to end anastomoses versus subclavian flap cohorts

Differences observed between limbs, between groups are reported in Table 2. The subclavian flap group had a significantly larger difference in lower arm muscle mass – right greater than left (94.5 ± 42.3 mls versus 37.7 ± 94.5 mls, $p = 0.008$), greater arm maximal cross-sectional area (right greater than left), (5.9 ± 2.8 cm^2 versus 2.9 ± 2.6 cm^2, $p = 0.038$). In the subclavian flap group, right versus left grip strength was also significantly greater than in the end to end anastomosis group (14.7 ± 8.3 versus 5.9 ± 5.3, $p = 0.016$) between the right and left arms.

Functional assessment

Functional dexterity test results between repair types are shown in Table 1. There were no other significant differences between the two groups.

Discussion

To our knowledge, this study provides the most complete structural and functional assessment of the upper limbs in adults who have undergone anatomically successful subclavian flap or end to end anastomosis for coarctation repair in childhood. We have found significantly greater right versus left arm muscle mass and grip strength differential in the subclavian flap repair patients, compared to the difference in right versus left arms in end to end anastomosis subjects.

The structural and functional consequences of left subclavian artery ligation in the treatment of congenital heart disease, whether it be Blalock Taussig shunt or subclavian flap repair, have predominantly been reported in paediatric populations.[7,9,20–22] Reduced blood flow at rest and during hyperaemia,[9] reduced longitudinal bone length [22,23] and muscle thickness, as measured by anthropometry, have been reported in subjects who have undergone subclavian flap repair.[20] Abnormalities in blood flow, arm length and grip strength have also been demonstrated in ipsilateral arm of patients with Blalock Taussig shunts.[21,24] However, these results have not been uniformly demonstrated, with Shenberger et al[7] finding no difference in blood flow nor static forearm strength, 9 years after subclavian flap repair.

In adults with repaired coarctation, evidence on the structural and functional sequelae of left subclavian ligation is scarce and

Table 1. Comparison of parameters in SF versus EEA.

	Total	EEA	Subclavian flap	p-value
Age, mean (SD)	38 ± 12	37 ± 14	38 ± 9	p = 0.839
Male gender, n (%)	14 (78)	6 (43)	8 (57)	p = 0.257
Mean systolic blood pressure right arm (mm Hg)	125 ± 13	129 ± 11	120 ± 13	p = 0.110
Mean systolic blood pressure left arm (mm Hg)	115 ± 16	123 ± 14	106 ± 13	p = 0.056
Mean diastolic blood pressure right arm (mm Hg)	80 ± 11	84 ± 10	75 ± 10	p = 0.017
Mean diastolic blood pressure left arm (mm Hg)	73 ± 13	79 ± 13	66 ± 9	p = 0.018
Height (cm)	174 ± 10.4	171 ± 11.0	176 ± 9.8	p = 0.368
Weight (kg)	75 ± 12.7	72 ± 20.6	78 ± 16.1	p = 0.545
Diabetes, n (%)	1 (6)	1 (100)	0	
Hypertension, n (%)	8 (44)	3 (38)	5 (63)	
Smoker, n (%)	5 (28)	2 (40)	3 (60)	
Hypercholesterolaemia, n (%)	2 (11)	1 (50)	1 (50)	
Bone metrics				
Right humeral length (mm)	31.9 ± 2.3	31.0 ± 1.4	32.9 ± 2.8	p = 0.079
Left humeral length (mm)	31.5± 2.4	30.7 ± 1.8	32.3 ± 2.8	p = 0.175
Right radial length (mm)	23.9 ± 1.8	23.3 ± 1.4	24.5 ± 2.0	p = 0.146
Left radial length (mm)	23.7 ± 1.8	23.1 ± 1.6	24.3 ± 1.9	p = 0.148
Right ulnar length (mm)	26.2 ± 1.8	25.6 ± 1.3	26.9 ± 2.1	p = 0.132
Left ulnar length (mm)	25.8 ± 1.7	25.3 ± 1.5	26.3 ± 1.9	p = 0.235
Muscle metrics				
Right upper arm volume (mls)	476.3 ± 140.8	440.2 ± 135.7	517.0 ± 143.9	p = 0.276
Left upper arm volume (mls)	437.1 ± 122.0	412 ± 117	464.7 ± 129.0	p = 0.396
Right upper arm MCSA (cm/2)	43.1 ± 13.2	39.0 ± 12.8	47.7 ± 12.8	p = 0.182
Left upper arm MCSA (cm/2)	40.4 ± 12.4	37.0 ± 12.0	44.1 ± 12.5	p = 0.248
Right lower arm volume (mls)	607.0 ± 183.9	556.0 ± 179.3	664.3 ± 183.0	p = 0.237
Left lower arm volume (mls)	542.6 ± 148.7	518.3 ± 157.9	569.8 ± 143.0	p = 0.598
Right lower arm MCSA (cm/2)	42.3 ± 12.1	39.0 ± 12.0	46.0 ± 11.8	p = 0.606
Left lower arm MCSA (cm/2)	38.0 ± 9.6	36.1 ± 9.9	40.1 ± 9.5	p = 0.666
Ultrasound metrics				
Pre-cuff flow mean (ml/min) - Right	100.4 ± 52.9	127.6 ± 56.3	73.2 ± 33.1	p = 0.024
Post-cuff flow mean (ml/min) - Right	378.4 ± 141.2	432.7 ± 168.9	323.8 ± 84.5	p = 0.110
Pre-cuff flow mean (ml/min) - Left	65.7 ± 33.8	69.8 ± 39.6	61.6 ± 28.6	p = 0.620
Post-cuff flow mean (ml/min) - Left	288.4 ± 66.2	299.6 ± 62.7	277.2 ± 71.6	p = 0.492
Baseline diameter right (mm)	3.9 ± 0.7	4.1 ± 0.9	3.6 ± 0.5	p = 0.134
Baseline diameter left (mm)	3.3 ± 0.4	3.4 ± 0.5	3.3 ± 0.4	p = 0.848
Percentage Change diameter right (mean ± SD)	6.1 ± 3.0	5.5 ± 3.1	6.7 ± 2.9	p = 0.422
Percentage Change diameter left (mean ± SD)	7.4 ± 3.7	7.7 ± 3.0	7.1 ± 4.4	p = 0.734
Functional metrics				
Purdue pegboard test - Right (mean score ± SD)	15.9 ± 1.4	15.7 ± 1.9	16.1 ± 0.7	p = 0.592
Purdue pegboard test - Left (mean score ± SD)	15.3 ± 1.8	14.7 ± 2.0	15.8 ± 1.6	p = 0.241
Purdue pegboard test - both (mean score ± SD)	13.7 ± 3.7	12.6 ± 2.0	14.7 ± 4.7	p = 0.228
Purdue pegboard test - Assembly (mean score ± SD)	35.3 ± 7.9	34.8 ± 8.8	35.7 ± 7.4	p = 0.832
Nine hole peg test - Right (mean score ± SD)	17.6 ± 1.6	17.8 ± 1.8	17.5 ± 1.4	p = 0.699

(*Continued*)

https://doi.org/10.1017/S1047951119000386 Published online by Cambridge University Press

Table 1. (*Continued*)

	Total	EEA	Subclavian flap	p-value
Nine hole peg test - Left (mean score ± SD)	18.1 ± 2.1	18.4 ± 2.6	17.7 ± 1.5	p = 0.467
QDASH (mean score ± SD)	6.7 ± 8.7	6.5 ± 5.1	6.8 ± 11.4	p = 0.949
Box Block right (mean score ± SD)	43.1 ± 14.4	39.4 ± 7.4	46.8 ± 18.9	p = 0.289
Box Block left (mean score ± SD)	42.9 ± 12.7	38.8 ± 6.6	47.1 ± 16.1	p = 0.171
JAMAR Grip Strength right (mean ± SD)	94.0 ± 29.0	82.5 ± 23.7	105.6 ± 28.4	p = 0.080
JAMAR Grip Strength left (mean ± SD)	83.7 ± 24.8	76.6 ± 24.4	90.8 ± 24.5	p = 0.236

MCSA = maximal cross-sectional area; DASH = disabilities of the arm, shoulder and hand; QDASH = quick DASH.

Table 2. Right versus left arm differences in structure and function by coarctation repair type.

	Total	EEA	Subclavian flap	P-value
Bone metrics				
Right versus Left difference in humeral length (mm)	0.4 ± 0.7	0.3 ± 0.7	0.7 ± 0.6	p = 0.221
Right versus Left difference in radial length (mm)	0.2 ± 0.4	0.7 ± 0.6	0.2 ± 0.4	p = 0.929
Right versus Left difference in ulnar length (mm)	0.4 ± 0.4	0.2 ± 0.4	0.6 ± 0.4	p = 0.155
Muscle metrics				
Right versus Left upper arm muscle volume difference (mls)	39.3 ± 42.3	27.7 ± 32.3	52.3 ± 50.3	p = 0.243
Right versus Left lower arm muscle volume difference (mls)	64.5 ± 46.9	37.7 ± 94.5	94.5 ± 42.5	p = 0.008
Right versus Left upper arm MCSA difference (cm/2)	2.7 ± 3.1	2.0 ± 3.1	3.6 ± 3.2	p = 0.311
Right versus Left lower arm MCSA difference (cm/2)	4.3 ± 3.0	2.9 ± 2.6	5.9 ± 2.8	p = 0.038
Ultrasound metrics				
Right versus left arm pre-cuff flow mean (ml/min) difference	31.7 ± 47.0	38.7 ± 45.7	24.6 ± 50.0	p = 0.543
Right versus left arm post-cuff flow mean (ml/min) difference	89.9 ± 132.0	133.2 ± 153.2	46.6 ± 96.7	p = 0.171
Right versus left arm pre to post cuff flow mean (ml/min) change	−55.2 ± 100.3	−75.4 ± 116.8	−35.0 ± 82.6	p = 0.409
Right versus left arm baseline artery diameter difference (mm)	0.5 ± 0.6	0.8 ± 0.8	0.3 ± 0.3	p = 0.110
Right versus left arm artery percentage change in diameter difference (%)	−1.3 ± 4.0	2.7 ± 2.9	−0.4 ± 5.0	p = 0.365
Functional Metrics				
Right versus left arm PPTscore difference	0.7 ± 1.5	1.0 ± 1.4	0.5 ± 1.7	p = 0.507
Right versus left arm NHP score difference	−0.4 ± 1.9	−0.7 ± 1.8	−0.2 ± 2.2	p = 0.628
Right versus left arm box score block difference	0.1 ± 2.8	0.6 ± 1.8	−0.3 ± 3.6	p = 0.501
Right versus left arm JAMAR grip strength difference (lbs)	10.3 ± 8.1	5.9 ± 5.3	14.7 ± 8.3	p = 0.016

MCSA = maximal cross-sectional area; PPT = purdue peg test; NHP = nine-hole peg test.

limited by subjectivity. Self-reported differences in arm length and muscle development post subclavian flap repair have been shown in some[2,6] but all studies.[1]

We found significant differences in right versus left lower arm muscle mass and grip strength of patients with subclavian flap repair compared to the differences seen in end to end anastomosis, whilst no significant differences were noted in the upper arm, suggesting a preferential effect on the left lower arm of ligation of the subclavian artery. This is in contrast to the findings of Van Son et al[9] who reported significant lower blood flow velocity, FMD, upper arm length and circumference in the left arm of subclavian flap repaired children (median age 8 years, IQR 2.5). These discordant findings likely relate to a number of factors. Our cohort's mean age (38 ± 12 years) is substantially older, and thus further from initial repair, and a reduction in blood flow abnormalities is known to occur with increased time from initial surgery,[21,22] presumably with the development of collateral blood supply. Therefore, it is unlikely time of initial repair had a significant impact. Collateral blood supply has also been demonstrated to be influenced by the location of subclavian artery division.[9] We did not assess the specific site of division on all our patients. Further, previous studies have utilised anthropometry for bone length and muscle circumference, where we utilised MRI for more accurate comparison of these parameters.

Our study reported higher diastolic blood pressure in end to end anastomoses patients when compared to the subclavian repair cohort and a non-significantly higher systolic blood pressure. This is in contrast to previous studies reporting higher blood pressures in subclavian flap repair cohorts.[25] The aetiology of late-onset hypertension in coarctation patients is thought to be multi-factorial including age of repair, type of repair, activation of the rennin–angiotensin system, residual coarctation, aortic arch type and intrinsic abnormalities in the aortic vasculature.[26] Our study was not sufficiently powered to detect meaningful differences nor causes of blood pressure differentials between groups.

We performed rigorous standardised functional testing of arm function and found no significant differences between subclavian flap repair and end to end anastomosis subjects when comparing differences between right arm and left arm function. This is in contrast to previous studies reporting subjective differences without any standardised objective testing.[2] It is likely that the lack of functional difference relates to learning and plasticity of motor skills but is possibly due to the insensitivity of tests applied, to pick up small functional differences between the right and left arm. Furthermore, arm function and dexterity are likely related to occupation and training effects, not only to anatomical and physiological structure.

Limitations

The assessment of upper limb muscle volume did not include the whole length humerus and shoulder, however the 15 cm proximal to the elbow joint did include the maximal biceps muscle volume and area. Blood flow was only assessed in one artery in each group and regional blood flow variations may exist and were not assessed. Left-handed subjects were not studied, and so our observations may not be generalisable to this group.

Conclusion

Subclavian Flap repair results in significant differences in muscle mass and grip strength between the right and left arms when compared to patients with end to end anastomosis in adults with repaired coarctation of the aorta.

Supplementary Material. To view supplementary material for this article, please visit https://doi.org/10.1017/S1047951119000386.

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Funding. This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Disclosures. The authors report no conflict of interest. These authors take responsibility for all aspects of the reliability and freedom from bias of the data presented and their discussed interpretation. The authors report no relationship to industry related to this study.

Ethics Approval. The study protocol was approved by the institutional review board of Royal Prince Alfred Hospital, Sydney Local Health District.

Contributors. All authors have read and approved the manuscript. None of them have declared conflict of interest. Each author has contributed significantly to the work.

Provenance and Peer Review. Not commissioned; externally peer reviewed.

References

1. Adams EE, Davidson WR, Jr., Swallow NA, et al. Long-term results of the subclavian flap repair for coarctation of the aorta in infants. World J Pediatr Congenit Heart Surg 2013; 4: 13–18.
2. Pandey R, Jackson M, Ajab S, Gladman G, Pozzi M. Subclavian flap repair: review of 399 patients at median follow-up of fourteen years. Ann Thorac Surg 2006; 81: 1420–1428.
3. St Louis JD, Harvey BA, Menk JS, O'Brien JE, Jr., Kochilas LK. Mortality and operative management for patients undergoing repair of coarctation of the aorta: a retrospective review of the pediatric cardiac care consortium. World J Pediatr Congenit Heart Surg 2015; 6: 431–437.
4. Cohen M, Fuster V, Steele PM, Driscoll D, McGoon DC. Coarctation of the aorta. Long-term follow-up and prediction of outcome after surgical correction. Circulation 1989; 80: 840–845.
5. Waldhausen JA, Petry EL, Kurlander GJ. Successful repair of subvalvular annular aneurysm of the left ventricle. N Engl J Med 1966; 275: 984–987.
6. Barreiro CJ, Ellison TA, Williams JA, et al. Subclavian flap aortoplasty: still a safe, reproducible, and effective treatment for infant coarctation. Eur J Cardiothorac Surg 2007; 31: 649–653.
7. Shenberger JS, Prophet SA, Waldhausen JA, Davidson WR, Jr., Sinoway LI. Left subclavian flap aortoplasty for coarctation of the aorta: effects on forearm vascular function and growth. J Am Coll Cardiol 1989; 14: 953–959.
8. Kir M, Ugurlu B, Unal N, et al. Revisiting subclavian flap repair for neonates and small infants. Pak J Med Sci 2015; 31: 131–135.
9. van Son JA, van Asten WN, van Lier HJ, et al. Detrimental sequelae on the hemodynamics of the upper left limb after subclavian flap angioplasty in infancy. Circulation 1990; 81: 996–1004.
10. Abe T, Loenneke JP. Handgrip strength dominance is associated with difference in forearm muscle size. J Phys Ther Sci 2015; 27: 2147–2149.
11. Skinner M. Standards for data collection from human skeletal remains. Jane E. Buikstra, Douglas H. Ubelaker. Q Rev Biol 1995; 70: 539–540.
12. Corretti MC, Anderson TJ, Benjamin EJ, et al. Guidelines for the ultrasound assessment of endothelial-dependent flow-mediated vasodilation of the brachial artery: a report of the International brachial artery reactivity task force. J Am Coll Cardiol 2002; 39: 257–265.
13. Roberts HC, Denison HJ, Martin HJ, et al. A review of the measurement of grip strength in clinical and epidemiological studies: towards a standardised approach. Age Ageing 2011; 40: 423–429.
14. Kellor M, Frost J, Silberberg N, Iversen I, Cummings R. Hand strength and dexterity. Am J Occup Ther 1971; 25: 77–83.
15. Oxford Grice K, Vogel KA, Le V, et al. Adult norms for a commercially available Nine Hole Peg Test for finger dexterity. Am J Occup Ther 2003; 57: 570–573.
16. Mathiowetz V, Volland G, Kashman N, Weber K. Adult norms for the Box and Block Test of manual dexterity. Am J Occup Ther 1985; 39: 386–391.
17. Kontson K, Marcus I, Myklebust B, Civillico E. Targeted box and blocks test: normative data and comparison to standard tests. PLoS One 2017; 12: e0177965.
18. Tiffin J, Asher EJ. The Purdue pegboard; norms and studies of reliability and validity. J Appl Psychol 1948; 32: 234–247.
19. Kennedy CA, Beaton DE, Smith P, et al. Measurement properties of the QuickDASH (disabilities of the arm, shoulder and hand) outcome measure and cross-cultural adaptations of the QuickDASH: a systematic review. Qual Life Res 2013; 22: 2509–2547.
20. Currarino G, Engle MA. The effects of ligation of the subclavian artery on the bones and soft tissues of the arms. J Pediatr 1965; 67: 808–811.
21. Skovranek J, Goetzova J, Samanek M. Changes in muscle blood flow and development of the arm following the Blalock-Taussig anastomosis. Cardiology 1976; 61: 131–137.
22. Todd PJ, Dangerfield PH, Hamilton DI, Wilkinson JL. Late effects on the left upper limb of subclavian flap aortoplasty. J Thorac Cardiovasc Surg 1983; 85: 678–681.
23. Lodge FA, Lamberti JJ, Goodman AH, et al. Vascular consequences of subclavian artery transection for the treatment of congenital heart disease. J Thorac Cardiovasc Surg 1983; 86: 18–23.
24. Harris AM, Segel N, Bishop JM. Blalock-Taussig anastomosis for tetralogy of Fallot. A ten-to-fifteen year follow-up. Br Heart J 1964; 26: 266–267.
25. Canniffe C, Ou P, Walsh K, Bonnet D, Celermajer D. Hypertension after repair of aortic coarctation–a systematic review. Int J Cardiol 2013; 167: 2456–2461.
26. Vigneswaran TV, Sinha MD, Valverde I, Simpson JM, Charakida M. Hypertension in coarctation of the aorta: challenges in diagnosis in children. Pediatr Cardiol 2018; 39: 1–10.