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Multimodality imaging in prenatal diagnosis and management of aortic arch anomalies

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ABSTRACT

There is a growing body of knowledge regarding the integration of multiple imaging modalities into the care of fetuses with aortic arch anomalies. This study aims to present our experience and fetal cardiac output data from a cohort of patients with discrete aortic coarctation and aortic arch hypoplasia.

Methods/Results: Retrospective review identified 25 fetuses with discrete aortic coarctation (n = 8) or aortic arch hypoplasia (n = 17) (with mean gestational age at diagnosis, 27.2 weeks; at birth 37.6 weeks). Mean combined ventricular output in all fetuses with normal ventriculo-arterial concordance was 383 ml/min/kg (median 362) with mean R:L cardiac output ratio of 2.83 (median 2.13). Of 23 neonates with primary surgical arch repair, all 7 with discrete coarctation underwent lateral thoracotomy without re-intervention. Of 16 with arch hypoplasia, 10 underwent primary lateral thoracotomy (3 with re-intervention) and 6 underwent primary sternotomy (1 with re-intervention). Of 2 with mild coarctation without primary repair, one with coexistent severe LV dysfunction underwent stenting and dilatation; the other is followed in the context of transposition of the great arteries status post arterial switch. Pre-op CT and 3D cardiac printing were obtained in 19 neonates. During follow-up, five (20%) had systemic hypertension.

Conclusion: Fetal R:L cardiac output ratio may provide complementary hemodynamic information in prenatal diagnosis of aortic arch anomalies. As expected, characterization of arch conformation remains critical for surgical planning. Pre-operative cardiac CT remains a useful adjunct to delineate arch anatomy for surgical planning purposes but may not reduce re-intervention by itself. Benefit of 3D cardiac printing remains difficult to quantify but has become an invaluable tool for family education. Based on our data, neonates with arch hypoplasia may benefit from midline sternotomy for more extensive surgical exposure and potentially reduced re-intervention rate.

1. Introduction

Coarctation of the aorta is a stenosis of the aortic arch, typically occurring distal to the origin of the left subclavian artery close to the patent or ligamentous ductus arteriosus. Usually a discrete narrowing, coarctation of the aorta falls on a spectrum with tubular aortic arch hypoplasia representing the extreme end [1]. Despite several recent studies aiming to improve prenatal detection, early diagnosis remains a challenge [2–6]. Early detection, appropriate parental counseling, and medical and surgical intervention, reduce morbidity and mortality [7]. Recent technological advancement with several imaging modalities including fetal echocardiography, transthoracic echocardiography, cardiac computed tomography (CT), and 3-dimensional (3D) cardiac printing provides an expanding capacity to characterize the aortic arch. This information can be integrated into more precise anatomic characterization, diagnosis, and management of aortic arch anomalies.

Currently the advantages and drawbacks of each modality are well-known [8–10]. To our knowledge, an analysis of their effect on the surgical approach and rate of re-intervention has not been clearly described in the literature. This study aims to present our experience and fetal cardiac output data from a cohort of patients with discrete aortic coarctation and aortic arch hypoplasia with the goal of adding to the growing body of knowledge regarding the integration of multiple imaging modalities into the care of these patients.

2. Methods

A retrospective review of the University of Minnesota Division of Pediatric Cardiology fetal database was performed and granted exempt status by the Institutional Review Board. Twenty-five fetuses with an aortic arch anomaly (excluding those with single ventricle physiology) were identified from the period between 2009 and 2015. Prenatal and

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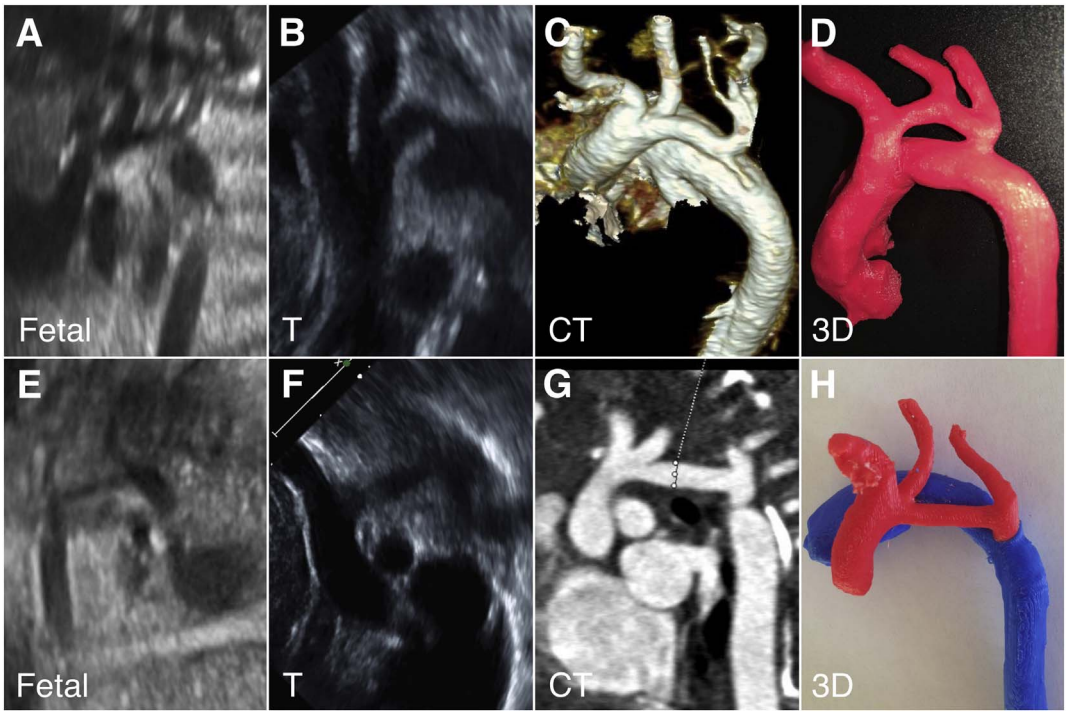

Fig. 1. Arch conformation (discrete coarctation vs. arch hypoplasia) as viewed on fetal echocardiogram (A vs. E), transthoracic echocardiogram (B vs. F), cardiac CT (C vs. G), and 3D cardiac print (D vs. H).

postnatal medical records were reviewed for gestational age, chromosomal abnormalities, and clinical course. Each fetal and transthoracic echocardiogram, cardiac CT, and 3D cardiac print imaging was reviewed and reanalyzed. These imaging studies were performed as part of clinical care.

Fetal and transthoracic echocardiography were performed using Philips iE33 or GE Voluson equipment. Each examination included 2-dimensional echocardiography with pulse, continuous wave, and color Doppler using variable frequency transducers. Cardiac CT imaging was performed using Siemens Flash CT scanner. 3D cardiac prints were produced with a Makerbot Replicator using cardiac CT data. Measurements on all images (excluding 3D-cardiac printing) were performed by one physician who was blinded to the final diagnosis and included aortic annulus diameter, pulmonary annulus diameter, carotid-subclavian index (ratio of the aortic arch diameter at the left subclavian artery to the distance between the left carotid artery and left subclavian artery), ascending aorta, transverse aorta, aortic isthmus, descending aorta, aortic velocity time interval, pulmonary velocity time interval, and heart rate. The anatomic diagnosis of coarctation versus arch hypoplasia (see Fig. 1) was defined qualitatively by the nature of the arch upon evaluation of echocardiographic and CT imaging.

The valve annuli were measured using the inner-to-inner edge. Velocity time interval measurements were made with an angle of insonation < 10–20° and were used in cardiac output calculations [11,12]. Fetal echocardiogram *Z*-scores were calculated based on gestational age [13,14]. Transthoracic echocardiogram *Z*-scores were calculated based on height and weight [15]. Cardiac CT Z-scores were calculated based on MR data, as there is no established cardiac CT Z-score data available [16]. Combined ventricular output and the distribution of right and left-sided output were calculated from fetal echocardiogram data and normalized by estimated fetal weight ($CO = HR * TVI * (d^2) * 0.785/wt.$ [cc/min/kg]) [11]. Acquired data was used in the comparison of patients with discrete coarctation and aortic arch hypoplasia.

2.1. Statistical Analysis

All data are presented as mean, median, and standard deviation. *Z*-scores for cardiac dimensions were calculated as above and a two-sample *t*-test for each variable was used to compare patients with discrete coarctation and aortic arch hypoplasia. All analyses were performed using R v3.3.1 with statistical significance defined as a $p < 0.05$ [17].

3. Results

Table 1 presents population characteristics of the 25 fetuses identified with an aortic arch anomaly. At the time of the first fetal echocardiogram, the mean gestational age was 27.2 weeks. At birth, the mean gestational age was 37.6 weeks and mean weight was 2.92 kg. At the time of primary surgical repair, the mean age was 16 days (median 9 days) and mean weight was 3.35 kg (excluding one patient repaired at two years of life).

Nine fetuses (36%) had an additional major cardiac anomaly including atrioventricular canal ($n = 4$), transposition of the great arteries ($n = 2$), double outlet right ventricle with normally related great arteries ($n = 2$), and left-ventricular non-compaction cardiomyopathy ($n = 1$). Twelve had a bicuspid aortic valve (48%) and one had a

Table 1
Population statistics.

Characteristic		Value
Total ($n = 25$)	Female	15
	Chromosomal anomaly	8
Mean [median] GA (weeks)	At diagnosis	27.2 [27.3]
	At birth	37.6 [38.3]
Mean [median] weight (kg)	At birth	2.92 [2.98]
	At primary repair	3.35 [2.89]*
Mean [median] age (d)	At primary repair	16 [6]*
	At re-intervention	221 [70]

* Excluding one patient repaired at two years of age.

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