Diagnostic Approach in Fetal Coarctation of the Aorta: A Cost-Utility Analysis

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Background: Coarctation of the aorta (CoA) is difficult to diagnose by fetal echocardiogram (F-Echo), often requiring multiple F-Echos during gestation and neonatal echocardiograms (N-Echos) after birth. Furthermore, CoA is the most common ductal-dependent lesion missed on routine physical exam.

Objectives: We sought to determine the most cost-effective diagnostic approach in caring for infants in whom an initial F-Echo is concerning for CoA.

Methods: Four paradigms for management after initial F-Echo could not rule out CoA were compared, with a single paradigm involving additional F-Echos: (1) multiple F-Echos for diagnostic clarity and performance of N-Echo on neonates with remaining high suspicion for CoA on F-Echos (prenatal-multiple), (2) no further F-Echo and performance of N-Echo on neonates with high suspicion for CoA on initial F-Echo (postnatal-selective), (3) no further F-Echo and performance of N-Echo on all neonates (postnatal-all), and (4) no further F-Echo or N-Echo with reliance on routine physical exam to identify afflicted infants (postnatal-none). Decision analysis models were constructed. Probabilities dictating clinical course and costs were calculated using our institution’s study population. The utility-state values were derived from existing literature. The measure of effectiveness was quality-adjusted life years. To represent societal perspectives, cost was defined as hospital reimbursement payments.

Results: From 2007 to 2014 at our institution, 92 patients were diagnosed with CoA and met the inclusion criteria for this study. These patients presented to care either through prenatal diagnosis (n = 31), postnatal examination findings while clinically well (n = 41), or after clinical deterioration in extremis (n = 20), with one patient subsequently dying. Presenting in extremis was associated with a 20% increase in the cost of their subsequent care and with a 51% increase in length of hospital stay. Postnatal-none was the least effective paradigm but also the least costly, thus forming the baseline model. Of the three other diagnostic approaches modeled, Postnatal-all was the cost-effective paradigm, maximizing utility due to avoidance of high-cost/low-utility disease states such as presentation in extremis and death. Prenatal-multiple was the next most effective but was also the most expensive.

Conclusions: Echocardiography is the screening gold standard in avoiding the devastating clinical manifestations of a missed CoA. When a diagnosis of CoA cannot be ruled out on initial F-Echo, the most cost-effective approach is performance of N-Echo on all neonates with no further prenatal evaluation. (J Am Soc Echocardiogr 2017; \( \cdot \) : \( \cdot \) : \( \cdot \) : \( \cdot \).)

Keywords: Aortic coarctation, Echocardiography, Prenatal, Cost-utility analysis

Prenatal diagnosis of congenital heart disease serves a critical role in parent education, prognostication, monitoring progression, and complication, as well as preparation for postnatal care. Coarctation of the aorta (CoA), which has a prevalence of four per 10,000 babies and represents 8% of congenital heart defects, is therefore a common perinatal concern among pediatric cardiologists.\(^1\)\(^2\) CoA is still the most common ductal-dependent cardiac defect missed at routine physical screening of the neonate; approximately 60%–80% of neonates with CoA remain unrecognized, presenting in low cardiac output states after spontaneous closure of the ductus arteriosus with resultant cardiovascular shock and death.\(^3\)\(^5\) Timely diagnosis and postnatal intervention are of critical importance as prenatal

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diagnosis of CoA has been shown to reduce morbidity and improve survival.\textsuperscript{7-11} However, CoA remains a difficult lesion to definitively diagnose by fetal echocardiogram (F-Echo) due to patency of the arterial duct and reliance on subjective measures such as ventricular asymmetry or dominance of the right heart structures. Ongoing efforts are being made to identify sensitive and specific quantitative prenatal echocardiographic markers, with most markers illustrating a poor positive predictive value.\textsuperscript{7-11} Still, there is a high false-positive rate for the fetal diagnosis of CoA, resulting in a large proportion of fetuses with multiple F-Echos during a pregnancy, initiation of prostaglandin E\textsubscript{1} administration, and postnatal management of fetuses with prenatal concern for CoA along with the costs associated with CUA. A decision tree was constructed to model four potential diagnostic approaches in a hypothetical patient population for the diagnosis of CoA. F-Echos after an initially inconclusive study may add expense but not add diagnostic clarity.\textsuperscript{7} Instead, the images may remain inconclusive, and providers must defer definitive diagnosis to the immediate postnatal period with acquisition of an N-Echo immediately after delivery. Little data are available to guide clinicians’ decisions in their diagnostic care of fetuses with prenatal concern for CoA and the aircapacity with which they should repeat F-Echos in lieu of deferring diagnosis until the acquisition of definitive N-Echo. Two recent studies employ cost-effectiveness analyses in understanding the utility of F-Echo in the diagnosis of congenital heart disease.\textsuperscript{12,13} We aimed to report on our single-center experience with prenatal evaluation and postnatal presentation of fetuses at risk for CoA along with the costs associated with their care. We hypothesized that a cost-utility analysis (CUA) could then be employed to improve the prenatal evaluation and postnatal management of fetuses with increased CoA risk by optimizing clinical outcomes while minimizing cost of care.

### Abbreviations

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<thead>
<tr>
<th>Abbreviation</th>
<th>Description</th>
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<tbody>
<tr>
<td>CoA</td>
<td>Coarctation of the aorta</td>
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<td>CPT</td>
<td>Common Procedural Terminology</td>
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<td>CUA</td>
<td>Cost-utility analysis</td>
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<td>EV</td>
<td>Expected value</td>
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<td>F-Echo</td>
<td>Fetal echocardiography</td>
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<td>ICER</td>
<td>Incremental cost-effectiveness ratio</td>
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<td>ICU</td>
<td>Intensive care unit</td>
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<td>LOS</td>
<td>Length of stay</td>
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<td>N-Echo</td>
<td>Neonatal echocardiography</td>
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<td>QALY</td>
<td>Quality-adjusted life years</td>
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**METHODS**

**Data and Assumptions**

A retrospective cohort study was performed and included all subjects at our institution between 2007 and 2014 with prenatal concern for CoA and/or who required surgical or transcatheter intervention for CoA in the first 30 days of life. The criteria for the prenatal diagnosis of CoA are both quantitative and qualitative. In our laboratory, we utilize aortic arch Z scores, flow acceleration through the arch, Doppler reversal of flow into the transverse arch, reversed foramen shunting, and novel measures as have been delineated in our laboratory.\textsuperscript{14-16} In addition, we look for subtle determinants of potential arch pathology including “splaying” of the distance between the left carotid and subclavian arteries, anatomic “tortuosity” of the aortic arch, and minor left heart pathology. In order to focus on a diagnosis of uncomplicated CoA, exclusion criteria included neonates with interventions for pathology unrelated to congenital heart disease during the hospitalization. Fetal subjects with no postnatal follow-up available were also excluded. Subjects in whom a prenatal diagnosis was made but whose postnatal evaluation revealed normal anatomy were only evaluated to quantify the cost of any postnatal hospitalization while verifying reassuring cardiac anatomy.

Subject records were reviewed to extract clinical variables. The term “extremis” was defined as initiation of inotropic support preoperatively in the first 24 hours of the hospitalization. Other cardiac diagnoses requiring intervention were defined as cardiac lesions that were intervened upon, either surgically or by catheter procedure during the first hospitalization. Aggregated costs were extracted from each individual’s medical record for the inpatient care beginning at their initial presentation and ending with subsequent discharge from that initial hospitalization. To capture societal expense, costs were defined as insurer reimbursement payments. Embedded in these hospitalization costs were all costs accrued during the initial hospitalization which would include preoperative intensive care unit (ICU) care, diagnostic imaging, surgical care, interventional catheter procedures, and the postoperative stay. The variable Standard Hospitalization represents the length of stay (LOS) and costs associated with those patients diagnosed prenatally or presenting clinically well. Extremis Hospitalization captures the experience and costs of those presenting in extremis. F-Echos with “high concern” were defined as those in which an N-Echo immediately after birth was advised. These data were aggregated and averaged to derive baseline assumptions for use in the CUA. Comparative statistics utilized a one-tailed Student’s t test. Seattle Children’s Hospital Institutional Review Board approved the study protocol.

To model prenatal costs that would not be captured in the hospitalization data, F-Echo costs were estimated using the sum of Common Procedural Terminology (CPT) codes 76825 (“Fetal echocardiogram 2D, complete”) and 76827 (“Doppler Echocardiogram, Fetal”). Similarly, each N-Echo cost was allocated costs associated with CPT code 93320 (“Echocardiogram 2D, complete”) and CPT code 93325 (“Doppler Echocardiogram, Complete”). Each F-Echo appointment was also allocated costs associated with CPT code 99245 (“Outpatient Consult Clinic Visit, Level 5”). Costs were reported in 2014 U.S. dollars, with a 3% discount rate to standardize values and were a mix of private and public insurer.

### Decision Analysis

The primary data from our institution were used to calculate cost and probability assumptions that were used in the decision tree analysis. Tree Age Pro 2014 (Williamstown, MA) was used in construction of the decision tool and performance of subsequent CUA. A decision tree was constructed to model four potential diagnostic approaches in a hypothetical patient population for whom initial obstetrical screening results in referral to a fetal cardiologist, and upon initial F-Echo the diagnosis of CoA remains equivocal (Figure 1). The first paradigm, prenatal-multiple, modeled the approach predominantly followed by this institution in which patients whose F-Echo was initially inconclusive were subject to two additional F-Echo studies later in gestation and those with “highly concerning” F-Echo studies were evaluated by N-Echo immediately after birth. The second paradigm, postnatal-selective, modeled the pursuit of no further F-Echos but the obtaining of N-Echos on those neonates with a “highly concerning” initial F-Echo. Postnatal-all was the third paradigm and modeled the decision to pursue no further F-Echos after an initially inconclusive study but to obtain N-Echos on all patients regardless of the magnitude of concern.
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