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Vascular rings and slings: Contemporary observations

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ABSTRACT

We reviewed our recent experience with vascular rings and slings, including observations from diagnostic imaging and surgery. We analyzed data from all patients diagnosed with a vascular ring or sling between September 2015 and August 2018. Our definition of vascular rings or slings did not include a left aortic arch with an aberrant right subclavian artery, accompanied by a left ductus arteriosus or ligamentum. We identified 74 patients for analysis. Of the 74, 45 (61%) had a right aortic arch with aberrant left subclavian artery and left-sided ductus arteriosus or ligamentum, 14 (19%) had a right aortic arch with a left-sided ductus or ligamentum and a normal left subclavian artery, 11 (15%) had a double aortic arch, 2 (3%) had a pulmonary artery sling, and 2 (3%) had a left aortic arch with aberrant right subclavian artery and a right-sided ductus arteriosus or ligamentum. Of the 74 patients, 40 (54%) were born in Nevada between January 2017 and August 2018. Of these 40, 33 (83%) were diagnosed prenatally. For the period January 2017 to August 2018, there were approximately 54,000 live births in Nevada. For right aortic arch versus double aortic vascular rings, 31 had a right aortic arch vascular ring for a prevalence of 5.7 per 10,000 live births, and 6 had a double aortic arch vascular ring for a prevalence of 1.1 per 10,000 live births ($p = 0.0001$). Vascular rings are common congenital cardiovascular malformations, especially right aortic arch vascular rings. Further, a high percentage of vascular rings can be detected prenatally.

1. Introduction

Past publications emphasize the rarity of vascular rings. Most quote a prevalence of < 1% of congenital heart disease, which approximates < 1 per 10,000 live births [1]. Nevertheless, for the general population of Southern Nevada, we previously reported a prevalence of 2.4 per 10,000 [2]. We attributed improved fetal detection as primarily contributing to the difference between our reported vascular ring prevalence and that of previous studies.

This study builds on our past vascular ring and right aortic arch publications [2–4]. We update our current data and discuss findings from computed tomography and surgery.

2. Methods

This investigation received approval from the local Institutional Review Board. We obtained data for this observational, non-randomized, retrospective study by inquiring our research database (Epi-InfoTM) and electronic health records. We analyzed data from patients, with a vascular ring or sling in situs solitus and levocardia, cared for at our center between September 2015 and August 2018. We excluded patients with dextrocardia or heterotaxic situs, including left and right isomerism and situs inversus. We defined vascular rings as vascular and or ligamentous structures encircling the trachea and esophagus, with or without symptoms. Despite previous reports [5], we do not consider a left aortic arch with an aberrant right subclavian artery and left-sided ductus arteriosus or ligamentum a vascular ring or a "partial" vascular ring, and we excluded these from analysis. The Children's Heart Center Nevada is the sole provider of congenital heart care in the state, and our database and electronic health records contain information on all patients diagnosed with a vascular ring or sling in Nevada.

A single radiologist, with 20-year's experience interpreting congenital cardiovascular malformations studies, including 12 years performing three-dimensional reconstructions from magnetic-resonance and computed-tomography imaging data sets, performed all studies

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analyzed. Computed tomography angiography was undertaken with a 256-multislice scanner (GE Revolution with three-dimensional processing on the AW VolumeShare 7 workstation, General Electric Healthcare, Chicago, Illinois, USA). Patients fasted 4–6 h pre-procedure. Infants from the neonatal intensive care unit were swaddled and soothed with Tootsweet™ (JLM Accutek Healthcare, Lane Cove, New South Wales, Australia). Infants beyond the neonatal period received intravenous sedation. In contrast to magnetic resonance imaging in which anesthesia is required for young children and infants, no patient required general anesthesia for computed tomography because of rapid image acquisition. Isovue 200 (Bracco Diagnostics, East Princeton, New Jersey, USA) at 2.0 mL/kg was used for enhancement. Average radiation dose was 0.5 mSv/kg (equivalent to 10–40 chest X-rays, patient dependent). Axial source images were used to observe for esophageal obliteration and proximal dilation. Axial source images were also used for tracheal cross-sectional area calculations and three-dimensional reconstructions. Tracheal cross-sectional area calculations were made proximal to and at the site of a vascular rings maximal effect. Neither bronchoscopy nor barium swallow procedures were included in routine vascular ring-sling workups.

We analyzed our prenatal detection rate for those born between January 2017 and August 2018. We undertook fetal cardiovascular evaluations as previously reported [6]. We defined a prenatal detection rate as the number of patients detected prenatally divided by the total number of patients identified born during the analysis period. For Nevada birth numbers, we inquired United States census information [7].

We used SPSS version 13.0 (SPSS Inc., Chicago, Illinois, USA) for data analysis. We used nonparametric testing for statistical analysis. We set a p-value of < 0.05 as significant.

3. Results

Table 1 summarizes data on all 74 patients including type of vascular ring or sling, age in months at presentation (for prenatal or neonatal detection we assigned a 0 month age); percent of females (predominantly female but not quite statistically significant at $p = 0.08$); those with syndromes (3 of which had chromosome 22q11.2 deletion syndrome); surgical intracardiac malformations; number repaired; and number and current age of those unrepaired in active follow-up.

Table 2 displays data from computed tomography imaging on a 30-patient cohort with right aortic arch vascular rings, in which imaging data sets were complete. For analysis, we divided the 30 patients into symptomatic versus asymptomatic at the time of imaging. Patients with vascular rings comprised of a right aortic arch with aberrant left subclavian artery and left-sided ductus arteriosus or ligamentum predominated. We evaluated percent tracheal cross-sectional area reduction for both types of right aortic arch vascular rings. For vascular rings with aberrant left subclavian arteries, symptomatic patients had a statistically significant higher percentage of tracheal cross-sectional area reduction at the site of the vascular ring than asymptomatic patients; however, there were too few patients with a normal left subclavian artery for a comparative analysis. In contrast, esophageal obliteration, accompanied by proximal esophageal distention, was common to both symptomatic and asymptomatic patients.

Table 3 presents surgical data for those followed a minimum of 6 months post-operative, demonstrating that all patients had symptoms related to the airway, gastrointestinal tract, or both at the time of vascular ring or sling surgery. We are cognizant that a prenatal diagnosis will draw special parenteral attention to symptoms; nevertheless, symptoms leading to surgery did often include early stridor and swallowing difficulties. All but one patient's symptoms resolved. Backer et al. previously reported the physiological importance of a Kommerell diverticulum to proximal left subclavian artery ratio of > 1.5 [8]; thus, for analysis, we divided the right aortic arch with aberrant left subclavian artery and left-sided ductus arteriosus or ligamentum vascular rings by this ratio. In the group with a ratio > 1.5, we resected the diverticulum and re-implanted the left subclavian onto the left carotid in all but one patient, with resolution of symptoms at follow-up. Those with a > 1.5 ratio were significantly older than those with a ratio $<$ 1.5. As the higher ratio patients presented at an older age, we cannot determine whether the ratio was high at birth or increased with time.

Fig. 1 is a Kaplan-Meier plot of 46 patients with a right aortic arch vascular ring, diagnosed prenatally or neonatally, which showed progressive loss of freedom from surgical intervention. Of these initial 46 patients, one was symptomatic as a neonate and underwent repair, 45 were asymptomatic as neonates; nevertheless, all those undergoing repair were symptomatic except for one patient that was recently electively operated at 36 months of age and lacked 6-month follow-up data.

Of the 74 total patients, 40 (54%) were born in Nevada between January 2017 and August 2018. Of these 40, 33 (83%) were diagnosed prenatally. There were no missed prenatal diagnoses. The 33 prenatal diagnoses were made from 3,102 pregnant women that were principally referred for fetal cardiac evaluation because of risk factors for fetal congenital heart disease or a suspected fetal cardiac malformation. For the period January 2017 to August 2018, there were approximately 54,000 live births in Nevada, for a total vascular ring-sling prevalence of 7.4 per 10,000 live births. Specifically, 31 had a right aortic arch vascular ring for a prevalence of 5.7 per 10,000 live births, and 6 had a double aortic arch vascular ring for a prevalence of 1.1 per 10,000 live births ($p = 0.0001$).

4. Discussion

This study is complementary to our previous reports and updates and expands our data analysis [2–4]. In total, vascular rings and slings are common cardiovascular malformations with a prevalence of 7.4 per 10,000 births in our population. Further, right aortic arch vascular rings are significantly more common than double aortic arches. Right aortic arch vascular rings are more common than an array of individual

Table 1
Summary data on all vascular rings and slings n = 74.

Type n (%)	Age presenting in months[a] median (range)	Female n (%)	Syndrome n (%)	Intracardiac malformation n (%)	Repaired n (%)	Unrepaired n (%) Current age in months median (range)
RAA ALS left PDA/lig 41 (55)	0 (0–180)	22 (54)	6 (15)	4 (10)	20 (49)	21 (51) 14 (0–24)
RAA normal LS left PDA/lig 18 (24)	0 (0–192)	12 (67)	0	2 (10)	4 (22)	14 (78) 9 (0 – 13)
DAA 11 (15)	0 (0–84)	7 (64)	0	0	10 (91)	1 (9) 9
PA sling 2 (3)	0	2 (100)	1 (50)	0	2 (100)	NA
LAA right DA/lig 2 (3)	2 (0–4)	2 (100)	1 (50)	0	2 (100)	NA

[a] Fetal/neonatal = 0 months, *ALS* aberrant left subclavian artery, *DAA* double aortic arch, *LAA* left aortic arch, *lig* ligamentum, *LS* left subclavian artery, *PA* pulmonary artery, *PDA* patent ductus arteriosus, *RAA* right aortic arch.

Table 2
Right aortic arch vascular ring computed tomography data n = 30.

	Symptoms at study n = 20	No symptoms at study n = 10	p
Age at study months median (range)	14 (0–168)	0 (0–9)	0.001
RAA ALS left PDA/lig n (%)	20 (100)	7 (70)	0.72
RAA normal LS left PDA/lig n (%)	0	3 (30)	0.19
% Tracheal reduction			
RAA ALS Left PDA/lig Median (range)	42 (0–78)	22 (0 – 33)	0.024
RAA Normal LS Left PDA/lig Median (range)	NA	30 (15–43)	NA
Esophageal obstruction & proximal distension n (%)	18 (90)	7 (70)	0.83

ALS aberrant left subclavian artery, *CT* computed tomography, *lig* ligamentum, *LS* left subclavian artery, *PDA* patent ductus arteriosus, *RAA* right aortic arch.

Table 3
Surgery & ≥ 6 months post-operative n = 26.

Type n	Prenatal DX n (%)	Symptoms pre-op n	KD resect + ALS RI n	Age @ Surg months median (range)	Symptoms post-op n
RAA ALS left PDA/lig					
KD/ALS > 1.5 n = 9	4 (44)	9	8	32 (5–168)*	0
KD/ALS < 1.5 n = 7	7 (100)	7	0	6 (0 – 20)*	1
DAA n = 7	4 (57)	7	0	2 (0 – 120)	0
PA sling n = 1	0	1	0	1	0
LAA right PDA/lig n = 2	0	2	0	3 (2,3)	0

ALS aberrant left subclavian artery, *DAA* double aortic arch, *DX* diagnosis, *KD* Kommerell diverticulum, *LAA* left aortic arch, *lig* ligamentum, *PA* pulmonary artery, *PDA* patent ductus arteriosus, *resect* resection, *RI* reimplantation, *surg* surgery.
* $p = 0.011$.

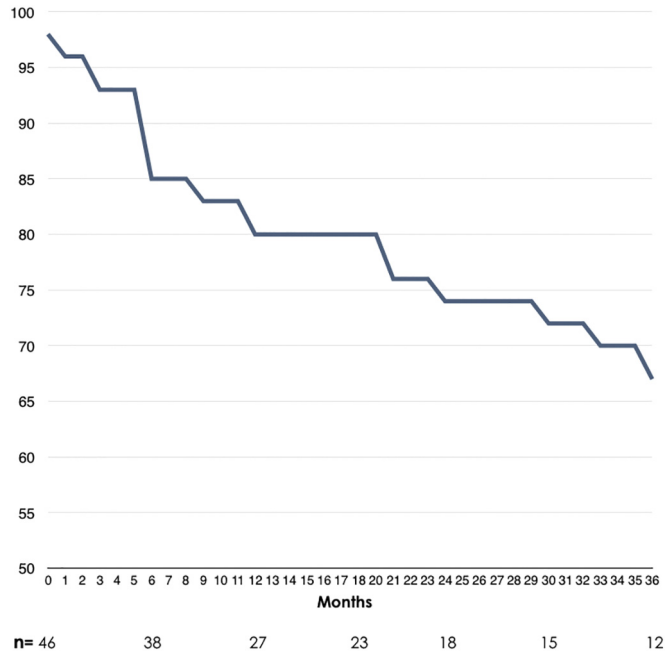

Fig. 1. Title: Kaplan Meier graph for right aortic arch vascular rings percent freedom from repair over time.

cardiovascular malformations that include hypoplastic left heart syndrome, transposition of the great arteries, aortic stenosis, coarctation of the aorta, atrioventricular septal defect, tetralogy of Fallot, and almost as common as pulmonary stenosis [9].

Improved prenatal detection has directly influenced the calculation of population-wide, vascular ring-sling prevalence figures in Nevada. Previously, we reported the results of our regionalized fetal cardiology program, which has improved the prenatal detection of significant congenital heart disease in our region [6]. Our approach consists of ongoing community-wide educational programs for general obstetric and specialized perinatal sonographers. Further, we provide on-site fetal cardiologists to supervise all diagnostic fetal echocardiograms at each of the region's perinatology clinics five days-a-week, complemented with 24/7 on-call services. Additionally, ultrasound equipment technology continues to improve, likely also contributing to an increasing rate of prenatal detection when compared to prior reports, including our previous experience [2,6]. Along with an improvement in detecting significant congenital heart disease, a secondary effect is an increase in prenatal detection of conditions such as vascular rings, primarily through the use of the three-vessel trachea view [10].

Some express caution over the prenatal detection of conditions asymptomatic at birth [11,12]. Savla and Weinberg wrote, “fetal diagnosis of a vascular ring should not be the only indication for surgery.” Additionally, Edwards and Hamilton's report equated an asymptomatic “loose vascular ring” and a persistent left superior a cava as “likely to be clinically insignificant” [12]. However, a persistent left superior vena cava is a normal variant and not equatable to a vascular ring, even a loose one. Neither an isolated fetal diagnosis of a vascular ring nor the presence of a loose vascular ring is by itself an indication for surgery. However, a Kaplan Meier analysis of right aortic arch vascular rings, primarily diagnosed prenatally and asymptomatic at birth, showed that within 36 months 33% had undergone surgery after patients developed symptoms. Also, the association between a Kommerell diverticulum and progressive proximal stenosis of an aberrant left subclavian artery can occur over time and may require intervention [13]. Surprisingly, undiagnosed vascular rings may also cause learning difficulties and adversely affect IQ [14,15]. Finally, the literature is replete with reports describing significant late gastrointestinal and respiratory complications, secondary to undiagnosed vascular rings. Undiagnosed vascular rings can present, even in the elderly, with cryptic histories and perplexing symptoms leading to protracted, traumatic, costly workups, and can occasionally result in mortality [16–23].

An alternative to concerns about prenatally diagnosed conditions asymptomatic at birth is that caution should be exercised over possible incongruities between subjective symptoms and objective signs. Even though we found a significantly greater reduction in tracheal cross-sectional area in those symptomatic versus asymptomatic (Table 2), there was some overlap in values. An overlap suggests symptoms or lack of symptoms may not be a deciding factor in every case, nor for that matter is objective cross-sectional values. Additionally, we also noted a higher degree of cross-sectional area reduction in older patients, data that at least raises the question whether these greater cross-sectional area reduction values were present at birth or worsened over time.

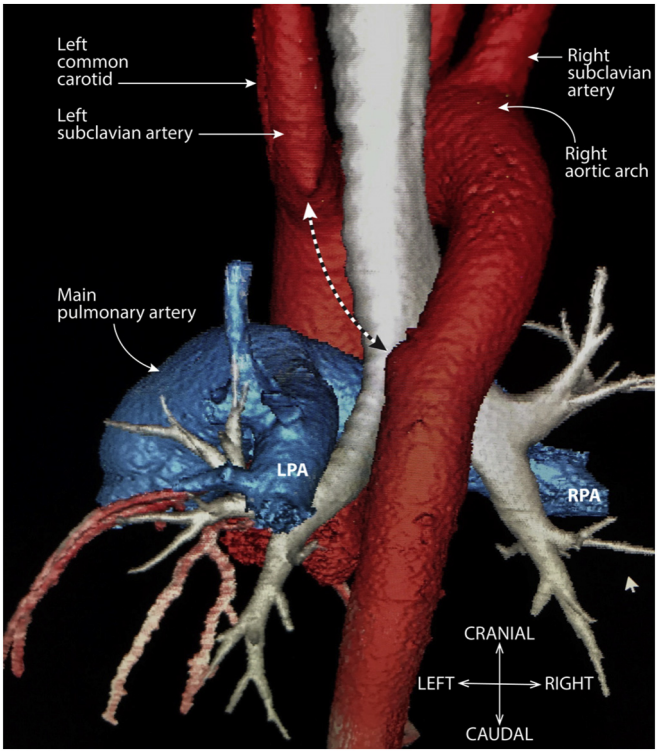

Fig. 2. Title: Double aortic arch.
Legend: Double headed arrow and dotted line identifies the location of the atretic left aortic arch, *LPA* left pulmonary artery, *RPA* right pulmonary artery.

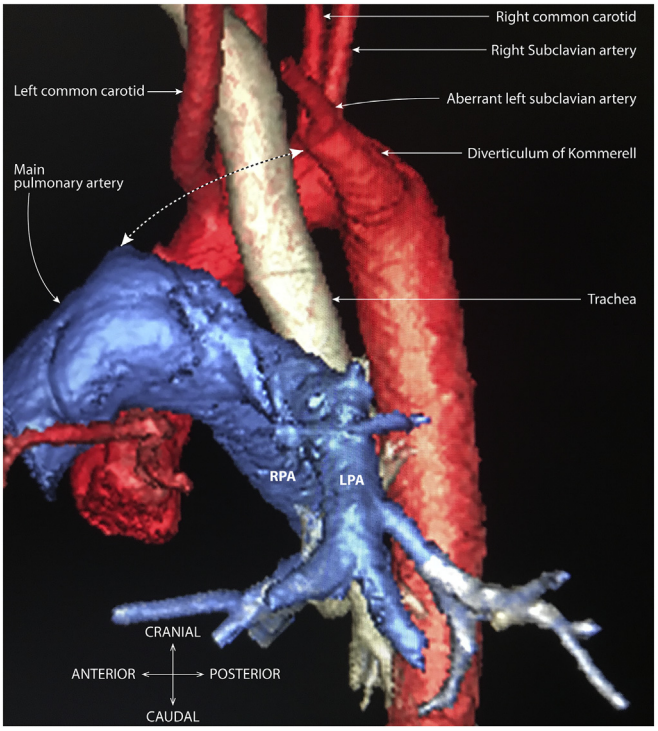

Fig. 3. Title: Right aortic arch vascular ring with aberrant left subclavian artery and left sided ligamentum.
Legend: Double headed arrow and dotted line indicate position of the ligamentum with minimal anterior deviation of the junction of the aberrant left subclavian and Kommerell diverticulum associated with minimal tracheal narrowing. *LPA* left pulmonary artery, *RPA* right pulmonary artery.

Further, from the Poiseuille equation, airway resistance is inversely proportional to the fourth power of the radius; thus, a 50% reduction in tracheal cross-sectional area is approximately proportional to a four-fold increase in airway resistance. Some have suggested discharge from follow up at between 2 and 4 years of age for those with an asymptomatic vascular ring [24,25]. However, our Kaplan Meier data and the suggestion that tracheal cross-sectional areas may progressively decrease over time both support the recommendation that asymptomatic patients should not be discharged from follow-up. Further, asymptomatic patients should, at some point, undergo computed tomographic imaging.

Muller et al. recently studied computed tomography versus bronchoscopy for evaluating the airway effects from a vascular ring, and they concluded that computed tomography was the preferable tool [26]. Computed tomography is excellent for defining the three-dimensional anatomical details. Even though we calculated airway cross-sectional area changes from axial source images, three-dimensional reconstruction can more clearly demonstrate subtle details such as nipple protrusions that, for example, can help define a double aortic arch's atretic left segment (Fig. 2). Additionally, another suggestive finding in right aortic arch vascular rings associated with an aberrant left subclavian artery is the approximate degree of anterior deviation at the junction of a ligamentum, Kommerell diverticulum, and origin of the aberrant left subclavian artery. Such deviations may relate to the tightness of the vascular ring, especially when coupled with the presence of external tracheal compression. Fig. 3 show less anterior deviation than Fig. 4, and the tracheal compression is greater in Fig. 4 versus Fig. 3.

This report's limitations include its retrospective nature and the limited number of patients for analysis. Further, we cannot account for asymptomatic, or symptomatic undiagnosed individuals. Without genetic testing on all patients, our incidence of genetic syndromes may be under-reported. Strengths include our high prenatal detection rates, robust database management, electronic health records on all

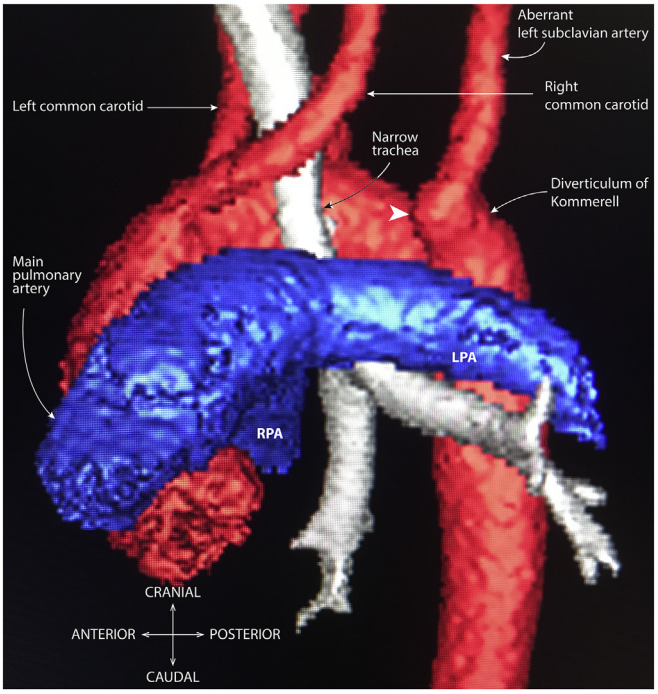

Fig. 4. Title: Right aortic arch vascular ring with aberrant left subclavian artery and left sided ligamentum.
Legend: Ligamentum not drawn in but significant anterior deviation of the junction (white arrowhead) of the aberrant left subclavian and Kommerell diverticulum associated with moderate tracheal narrowing. *LPA* left pulmonary artery, *RPA* right pulmonary artery.

diagnosed patients from the state of Nevada, and avoidance of relying on third-party sources.

In conclusion, vascular rings are common congenital cardiovascular malformations, especially right aortic arch vascular rings. Further, a high percentage of vascular rings can be detected prenatally. All symptomatic patients should undergo surgical repair. In those with a Kommerell diverticulum to aberrant left subclavian artery ratio of > 1.5, repair should likely be accompanied by diverticulum resection and reimplantation of the aberrant left subclavian artery. Asymptomatic patients should be routinely followed and observed for the development of symptoms and then referred for surgical repair. Genetic testing should be considered, especially for those with a right aortic arch vascular ring associated with an aberrant left subclavian artery. Finally, we recommend that patients with vascular rings, even if asymptomatic, not be discharged from follow-up, as a vascular ring represents pathology and numerous studies show late presentation of symptomatic patients.

Declaration of Conflicting Interests

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