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Unusual morphology of an aberrant right subclavian artery: a case report and systematic review of cadaveric studies

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ABSTRACT

Background: An aberrant right subclavian artery (ARSA) is a rare vascular anomaly defined by the origin of the right subclavian artery from the aorta, distal to the left subclavian artery (LSA). An ARSA was found in an 83-year-old female during cadaveric dissection for anatomy teaching, which had an unusually narrow lumen compared to previously reported cases.

Materials and methods: The specific morphology of this ARSA variant was characterised by further dissection and measurements. A systematic review of cadaveric case reports with quantitative evaluation of ARSA dimensions was conducted for comparison with this case.

Results: This ARSA variant exhibited an unusually narrow lumen (8.22 mm^2) compared to the LSA (152.55^2), with an ARSA: LSA ratio of 0.24. The systematic review included 17 studies with 23 cases of ARSAs. Of these cases, 20 contained details of ARSA diameter, 19 of which were wider than in this case. In all studies where both ARSA and LSA diameters were measured, ARSA was found to be significantly wider than LSA (mean ratio of ARSA to LSA: 1.49, range: 1.09–2.00). Several other variations were reported in these studies, such as concomitant aortic arch branch anomalies and differing origins of ARSA as defined by vertebral level and relation to the aortic arch.

Conclusions: This case report presents an unusually narrow ARSA which has not been previously described in the literature. As there is limited evidence on how ARSA morphology affects clinical outcomes, further research is needed to better inform management of ARSAs.

Keywords: aberrant right subclavian artery, arteria lusoria, morphology, dysphagia, subclavian steal

Introduction

An aberrant right subclavian artery (ARSA), also known as arteria lusoria, is a congenital anomaly where the right subclavian artery arises directly from the aortic arch or proximal descending aorta instead of the brachiocephalic trunk [27]. The ARSA variation has a reported incidence of 0.4–1.8%, with a female preponderance [32, 38]. The anomaly results from the abnormal regression of the 4th right vascular arch and proximal right dorsal aorta, with persistence of the 7th intersegmental artery [7, 38]. In-utero, the presence of an ARSA through foetal ultrasonography has been shown to be a risk marker for Down syndrome and this finding should prompt consideration for further chromosomal abnormalities [43]. Most ARSAs have a retro-oesophageal course (80%), though the artery may pass between the trachea and oesophagus (15%) or anterior to the trachea (5%) [32].

Several other anomalies can coexist with an ARSA, including a bicarotid trunk (BCT), Kommerell's diverticulum (KD), aneurysms, and constrictions of the aberrant artery along its course [34]. While an ARSA can be associated with different clinical manifestations, around 90% of cases are asymptomatic. In symptomatic cases, the ARSA typically affects swallowing (dysphagia) or the upper respiratory tract (dyspnoea and/or cough) due to oesophageal or tracheal compression [38]. Individuals with an ARSA may also be subject to subclavian steal, with symptoms of vertebrobasilar insufficiency [22].

This study reports our findings of an unusually narrow ARSA compared to previously reported cadaveric studies and reviews the evidence regarding the impact of size and morphology of ARSAs on the symptomatology of affected patients.

Materials and methods

During routine cadaveric dissection in the Human Anatomy Centre, Department of Physiology, Development and Neuroscience at the University of Cambridge, an aberrant right subclavian artery (ARSA) was noted in an 83-year-old female who had died of an intracranial haemorrhage. The donor had provided advanced consent for the use of her body for

anatomical teaching and research after death, in compliance with the Human Tissue Act 2004. The donor had been prepared using an arterial embalming technique by cannulation of the right common carotid artery and injection under pressure of a solution containing 38% ethanol, 1.5% methanol, 4.2% formaldehyde and 56.3% distilled water.

After further careful dissection of the aortic arch and its direct branches, the morphology of the aortic arch anomaly was examined and photographed *in situ*. The aortic arch was then explanted for detailed investigation. The following definitions were used for consistency and clarity. The “proximal” ARSA/LSA describes the part of the artery from its aortic origin to its first branch, the vertebral artery (VA). The “distal” ARSA/LSA describes the part of the artery beyond the respective vertebral artery, for which measurements were taken at the origin and at two further points, 2 cm and 4 cm distant from the origin.

Measurements of external vascular dimensions included: the right common carotid artery (RCCA), left common carotid artery (LCCA), left subclavian artery (LSA) and the ARSA. Measurements were also taken of the right (RVA) and left (LVA) vertebral arteries, the lengths of the proximal ARSA and LSA, and the dimensions of the distal ARSA and LSA. Two authors (PM and SF) used Mitutoyo digital callipers (to 2 decimal places) to independently measure the maximum diameter of the vessel and the diameter perpendicular to it at specific points. Each measurement was taken twice by each author, with a mean value established. From these values, the cross-sectional area was calculated using the formula for the area of an ellipse, πab, and the approximate perimeter $\pi(a + b)$ ($a = ½$ maximum diameter, $b = ½$ perpendicular diameter).

The unusual morphology in our specimen prompted a systematic review of the sizes of other cadaveric ARSAs documented in the literature. Using the key words of "aberrant right subclavian artery" OR “arteria lusoria” AND “cadaver”, we searched the databases of MEDLINE via PUBMED and Google Scholar on 16th May 2023. Criteria for inclusion were 1) English language studies 2) Human cadaveric/post-mortem studies and 3) Quantitative evaluation of ARSA +/– other aortic arch branches. Our methods supported an in-depth analysis [50] to provide a comprehensive study into this well-known but relatively rare vascular anomaly.

Results

Our case showed an anomalous left-sided aortic arch morphology which lacked a brachiocephalic trunk (BCT) and gave the following direct branches from proximal to distal: RCCA (36.1 mm^2), LCCA (79.9 mm^2) and LSA (152.5 mm^2). Distal to the LSA, the ARSA

(8.20 mm^2) arose from the medial aspect of the descending aorta at approximately the level of the fifth thoracic vertebra. It ascended superomedially at an acute angle posterior to the oesophagus towards the thoracic aperture (Fig. 1).

This proximal part of the ARSA (length 8.30 cm) had a narrow calibre (origin: 8.20 mm^2, midpoint 7.93 mm^2), until approximately 0.90 cm before giving its first branch, the RVA, where it began to enlarge (endpoint: 26.0 mm^2) and pass laterally inferior to the clavicle to the right upper limb. Distal to the origin of the RVA, the cross-sectional area of the ARSA (27.5 mm^2 to 38.3 mm^2) was smaller than that of the distal LSA (36.9 mm^2 to 42.8 mm^2). The RVA (25.8 mm^2) was also narrower than the LVA (45.4 mm^2), both of which were the first branch of their respective subclavian arteries. A summary of these external vascular dimensions is presented in Table 1. The ARSA was opened to show a narrow, but patent, lumen (Fig. 2).

In our systematic review, the initial database search yielded 293 abstracts. Of these, 72 abstracts were selected for in-depth review, with 17 papers eventually included in the study. The 17 studies included 23 ARSAs of varying morphology. Cadaver sex was available in all cases, with the age and ethnicity available in only 13 and 12 cases, respectively. All but one of the cases in the review were retro-oesophageal. Where available, the vertebral level of each ARSA origin and the presence of concomitant vascular anomalies were recorded.

Twenty ARSAs were evaluated using diameter, two by cross-sectional area and one by perimeter. Where diameter was used, the mean ARSA diameter was 12.22 mm (range 1.91–17.30 mm). Of these 20 ARSAs, 19 reported measurements that were larger than in our variant. Of the 11 cases which reported LSA diameter (mean: 9.09 mm, range 1.68–13.80 mm), all were smaller than in our study (15.45 mm).

We further examined the relationship between ARSA and LSA. All 13 cases with measurements of both ARSA and LSA described an ARSA larger than its respective LSA (ARSA > LSA), in contrast to our study (LSA > ARSA), seen in Figure 3A. In the 11 cases which reported both LSA and ARSA diameter, the ratio of ARSA to LSA was 1.49 (range: 1.09–2.00), in contrast to our ratio of 0.24, seen in Figure 3B.

Summary of ARSA dimensions systematic review is seen in Supplementary Table 1.

Discussion

An ARSA is symptomatic in only 7–10% of those with the anomaly [10]. First described by Bayford as “dysphagia lusoria” in 1787, dysphagia is the most common symptom associated with ARSA, occurring in 70% of symptomatic individuals [3, 24]. Dysphagia can occur if the

course of the aberrant artery leads to oesophageal compression and has been shown to be more likely in cases with a shorter median distance between the ARSA and the trachea, as well as in cases with a higher median luminal area [24]. Adult patients are most likely to present with dysphagia [38], with potential causative mechanisms being increasing rigidity in the oesophagus [46], increased tracheal rigidity [23], Kommerell's diverticulum (KD), formation of an aneurysm [18, 25], elongation of the aorta [48] or atherosclerotic changes in the wall of the ARSA [38]. Other symptoms associated with ARSA include dyspnoea, pain, cough and weight loss, which are similarly attributed to compression of adjacent structures [38].

Other vascular abnormalities often accompany ARSAs. According to a review conducted by Polguj et al. [38] on 15 cadaveric and 126 clinically documented cases, the most frequently observed vascular abnormalities were bicarotid trunk (BCT; 19.2%), KD (14.9%), aneurysm (12.8%) or right-sided aortic arch (9.2%). In our review of cadaveric studies only, BCT was explicitly mentioned in 6/24 (25%) cases, while KD was mentioned in 3/24 (12.5%) cases. Of these associated vascular abnormalities, most of the literature discussing symptomatic ARSA presentation has focussed on compression of adjacent structures such as the trachea or oesophagus [8, 24].

The unique morphology in this case led us to consider the potential consequences of such a narrow ARSA lumen. One presentation could be subclavian steal syndrome (SSS); which occurs when retrograde blood flow through the right vertebral artery causes vertebrobasilar insufficiency with symptoms such as diplopia, vertigo and presyncope [21, 22]. In comparison to KD, SSS is less well documented [22]. The sudden increase in ARSA calibre just after the RVA branches may suggest that vascular compensatory sequalae were present, possibly due to retrograde blood flow from the vertebrobasilar circulation.

Furthermore, whilst this ARSA variant had an origin that was more distal in comparison with the ARSAs reported elsewhere, there is scarce literature on the effect of the ARSA origin on the symptomatology of affected patients [7]. There is no clear association between the vascular anomaly present and the origin of the ARSA in relation to vertebral level or the course of the artery [7]. From its relatively distal origin, the course of the ARSA in our study ascended almost parallel and adjacent to the oesophagus and trachea. This contrasts with cases displaying a proximal origin from the aortic arch, where the ARSA crosses these structures in a perpendicular orientation, which could theoretically pose a greater risk of extrinsic compression and subsequent symptoms. The origin of the ARSA may also affect the approach, success, and complication rates of coronary angiography [49].

Symptomatic ARSAs typically require surgical treatment [17, 51], while asymptomatic patients may be treated clinically by controlling hypertension, as well as with platelet aggregation inhibitors and cholesterol reducers as secondary measures [33]. The vascular sequalae of asymptomatic ARSAs may necessitate surgical management, with the aim of preventing or treating complications such as aortic dissection, concomitant aortic aneurysmal disease, or an enlarging KD [11]. Moreover, even in the absence of symptoms, an ARSA may present a challenge in procedures such as oesophagectomies [27]. Additional studies on asymptomatic patients with KD would be useful to determine specific size criteria for operative procedures [11].

The open surgical interventional techniques used to treat ARSAs are arterial reimplantation/transposition or bypass. For many years, this necessitated a left-sided thoracotomy with division and ligation of the ARSA [15, 52]. Endovascular techniques are increasingly popular [11], but prerequisites for such procedures include sufficient size of the access arteries, limited tortuosity, and suitable proximal and distal neck morphology [51]. More recently, a hybrid approach has been used in which an open procedure may follow the endovascular repair to perform further vascular reconstruction. This approach is proving successful in the short term. However, long-term follow-up is needed to monitor potential complications and durability of the interventions [26, 45]. The safety and efficacy of these three treatment approaches are comparable and provide satisfactory outcomes to improve or eradicate symptoms [26].

Conclusions

This case report describes an unusually narrow ARSA, with a systematic review of other cadaveric studies and comparative analysis. While the literature suggests that the size of an ARSA may directly correlate with an increased risk of symptoms, there is little evidence regarding the effect of other anatomical features, such as ARSA origin, on clinical presentation and subsequent management. Further characterisation of ARSA morphology by radiological studies may help predict symptomatology and in doing so, provide further insights in how best to treat the sequelae of ARSAs.

ARTICLE INFORMATION AND DECLARATIONS

Ethics statement

The donor had provided advanced consent for the use of her body for anatomical teaching and research after death, in compliance with the Human Tissue Act 2004.

Author contributions

Kseniia Panteleeva — data analysis, interpretation, writing (majority). Dalia Bornstein — writing*. Neel Badhe — writing*. Perry Maskell — conception, design, data collection, writing. Cecilia Brassett — writing/editing. Sarah Fawcett — editing, data collection. *DB and NB equal contribution

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Conflict of interest

The authors have no conflict of interest to declare.

Supplementary material

The supplementary material contains:

- **Supplementary Table 1.** Summary of ARSA dimensions systematic review.

and it is available on Journal's website.

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Table 1. External dimensions of the aortic arch and other relevant arteries.

Vessel	**Cross-sectional area [mm^2]**
Ascending aorta origin	632.1
Descending aorta (T4 level)	370.7
Right common carotid artery origin	36.1
Left common carotid artery origin	79.9
Left subclavian artery (proximal) (length 3.2 cm) L	Origin: 152.5 Endpoint: 115.3
Left subclavian artery (distal)	Origin: 38.3 2 cm point: 36.9 4 cm point: 42.8
Aberrant right subclavian artery (proximal) (length 8.3 cm)	Origin: 8.20 Midpoint: 7.93 Endpoint: 26.0
Aberrant right subclavian artery (distal)	Origin: 38.3 2 cm point: 27.5 4 cm point: 32.2
Right vertebral artery	25.8
Left vertebral artery	45.4

“Proximal” denotes the subclavian artery from its origin at the aorta to the origin of the respective vertebral artery. “Distal” denotes the subclavian artery distal to the origin of the respective vertebral artery. In the sequential measurements of the distal subclavian arteries, “2 cm” and “4 cm” denote the cross-sectional area of the arteries at 2 cm and 4 cm distal to the respective vertebral artery.

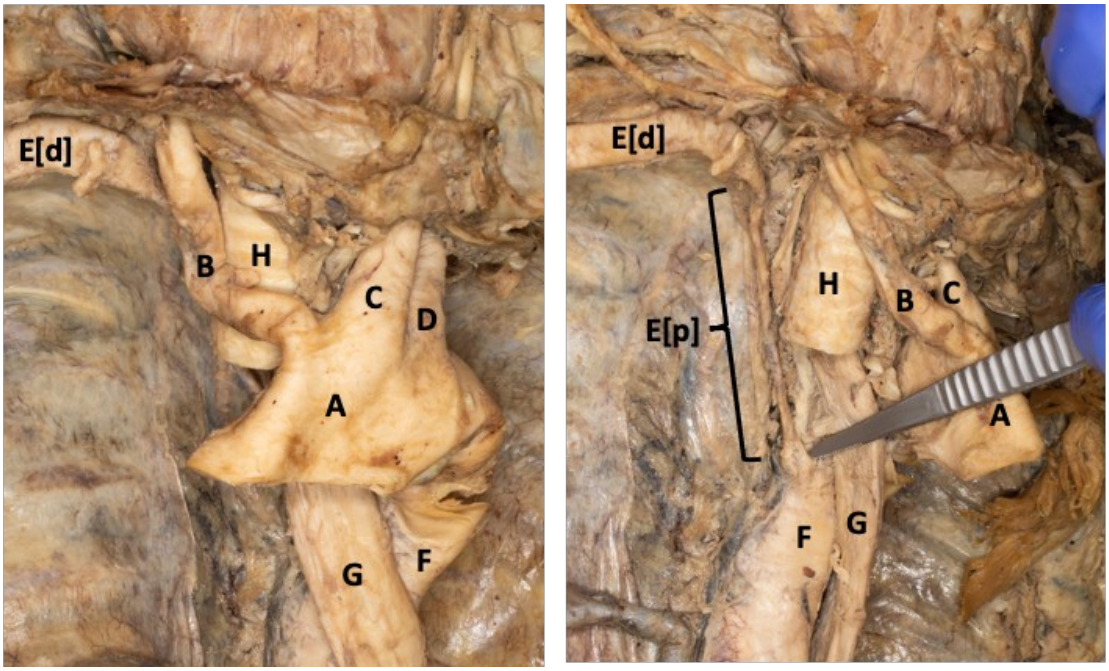

Figure 1. The aberrant right subclavian artery in situ (left) with surrounding structures retracted (right). A — aortic arch, B — right common carotid, C — left common carotid, D — left subclavian artery, E — aberrant right subclavian artery: p — proximal, d — distal; F — thoracic aorta, G — oesophagus, H ─ trachea.

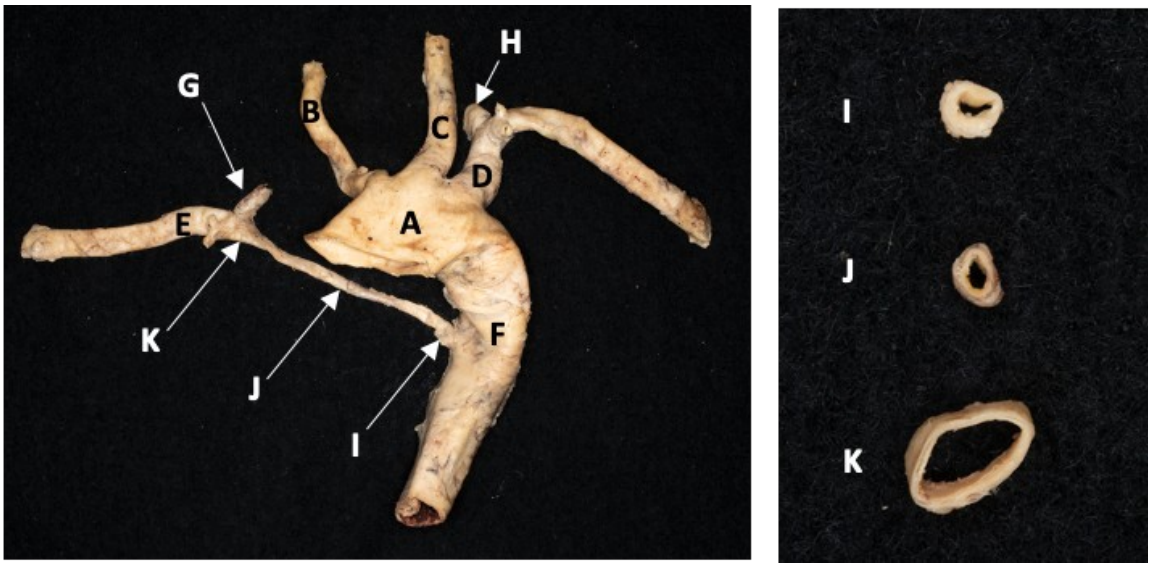

Figure 2. Explanted cadaveric aortic arch (left) with aberrant right subclavian artery cross sections (right). A — aortic arch, B — right common carotid, C — left common carotid, D — left subclavian artery, E — ARSA, F — thoracic aorta, right G — vertebral artery, H — left vertebral artery, I — ARSA origin, J — proximal ARSA midpoint, K — proximal ARSA endpoint. ARSA — aberrant right subclavian artery.

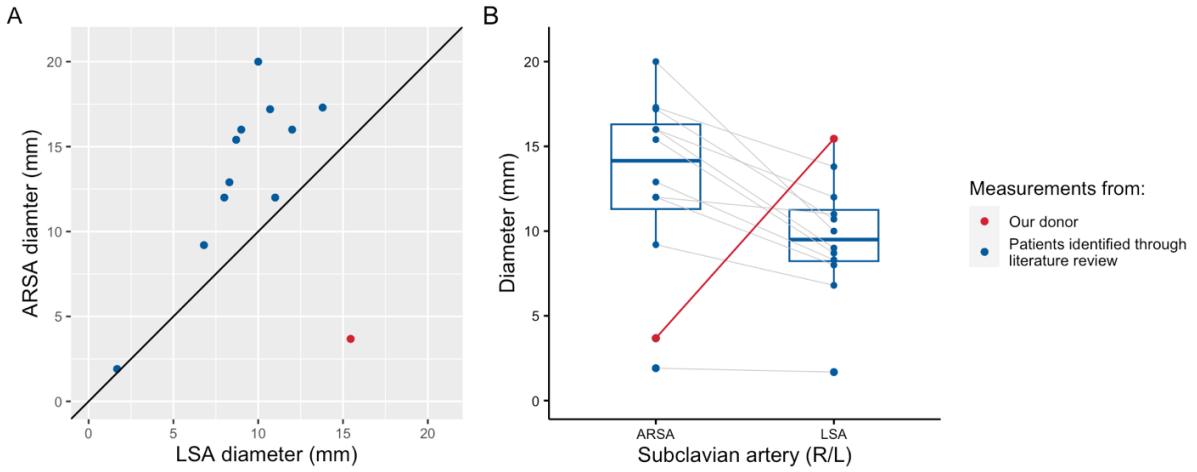

Figure 3. Scatter plot (**A**) and boxplot (**B**) of data from the systematic review of reported aberrant right subclavian artery (ARSA) diameter compared against left subclavian artery (LSA) diameter.

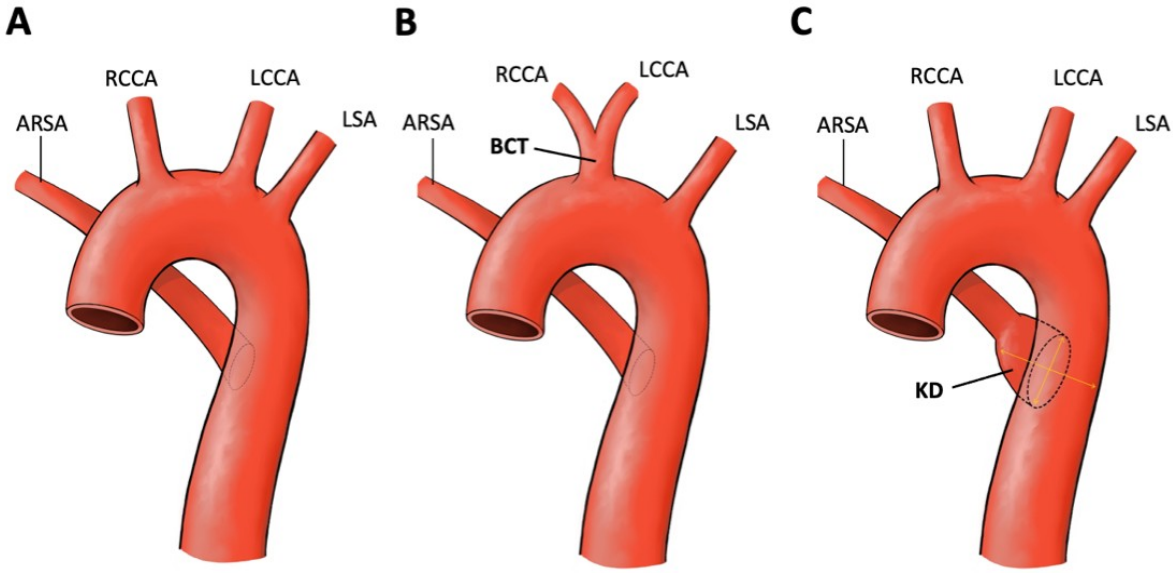

Figure 4. Common vascular abnormalities occurring with an aberrant right subclavian artery. **A.** ARSA without co-occurring vascular abnormalities; **B.** ARSA and bicarotid trunk; **C.** ARSA with Kommerell's diverticulum. ARSA — aberrant right subclavian artery; BCT — bicarotid trunk; KD — Kommerell's diverticulum; LCCA — left common carotid artery; LSA — left subclavian artery; LVA — left vertebral artery; RCCA — right common carotid artery; RVA — right vertebral artery.

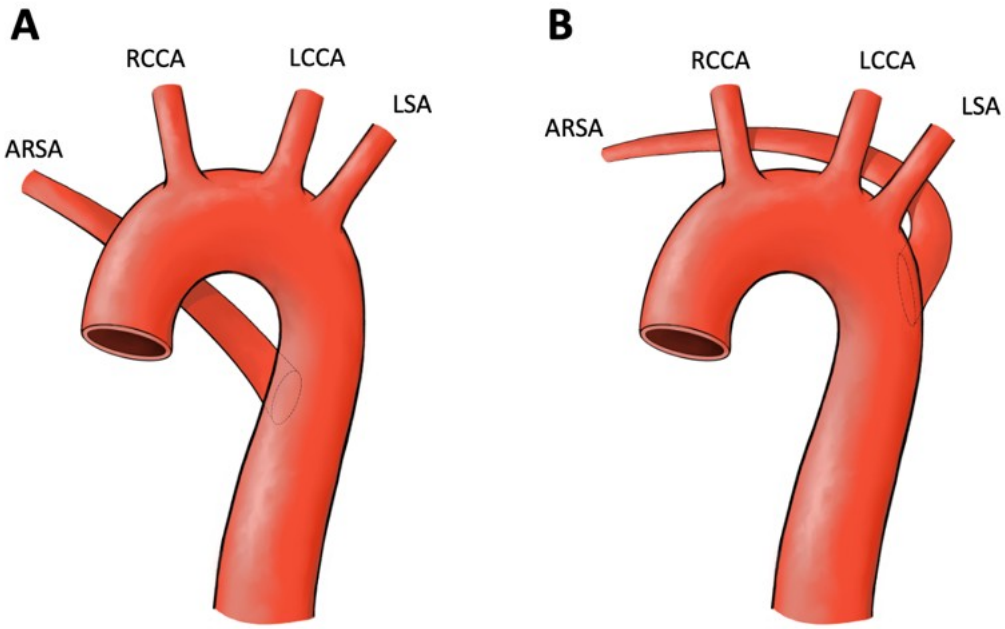

Figure 5. Documented aberrant right subclavian origin and position variants. **A.** ARSA originating from the medial/inside border of the descending aorta; **B.** ARSA originating from the posterior aortic arch. ARSA — aberrant right subclavian artery; LCCA — left common carotid artery; LSA — left subclavian artery; RCCA — right common carotid artery.

Supplementary Table 1. Summary of ARSA dimensions systematic review.

Study [Ref]	Cases	Cadaverdemographics (sex, age, ethnicity)	ARSA Diameter [mm]	ARSA Area [mm^2]	ARSA perimeter [mm]	LSA comparison	Origin location	Course	Other aortic arch anomalies present
Present study	1	Female, 83, Caucasian	3.68	8.2	10.16	LSA diameter 15.45 mm, LSA Area 152.5 mm2	Descending Aorta — T5	Retro–oesophageal	None
Alghamdi et al., 2020, Saudi Arabia [1]	1	Female, unknown, unspecified	16	–	–	LSA diameter 9 mm	Aortic arch	Retro-oesophageal	KD
Almenar-García et al., 2002, Spain [2]	1	Male, 82, unspecified	12	–	–	NA	Aortic arch — T3	Retro-oesophageal	NS
Buffoli et al., 2021, Italy[6]	1	Female, unspecified, Caucasian	9.2	–	–	LSA diameter 6.8 mm	Aortic arch	Retro-oesophageal	BCT, ectopic vertebral artery origin
Esumi et al., 2023,	1	Male, 62, Japanese	20	–	–	LSA diameter	Aortic arch — T3	Retro-oesoph	NS

Japan [13]						10 mm		ageal	
Fazan et al., 2003, Brazil [14]	1	Female, 54, Caucasian	16	–	–	LSA diameter 12 mm	Aortic arch	Retro-oesoph ageal	NS
Kaidoh et al., 2011, Japan [20]	1	Female, 75, Japanese	8	–	–	NA	Aortic arch — T3 (3 cm below the origin of the LSA)	Retro-oesoph ageal	KD
Makgalwa et al., 2007, South Africa [28]	1	Female, 44, black	17.3	–	–	LSA diameter 13.8 mm	Aortic arch (3 mm distal to LSA)	Retro-oesoph ageal	NS
Mirande et al., 2020, United States [29]	2	Male, 63, Caucasian	–	482.7	–	LSA area 142.3 mm^2	Aortic arch	Retro-oesoph ageal	NS
		Female, 73, Caucasian	–	305.7	–	LSA area 78.17 mm2	Aortic arch (posterior surface)	Retro-oesoph ageal	NS
Namking et al., 2009, Thailand [31]	1	Female, 80, unspecified	15.4	–	–	LSA diameter 8.7 mm	Aortic arch (crosses midline at T3)	Retro-oesoph ageal	NS
Natsis et al., 2011,	2	Male, 76, Caucasian	12	–	–	NA	Aortic arch — T4	Retro-tracheal	BCT

Greece [35]		Male, 81, Caucasian	16	–	–	NA	Aortic arch — T3	Retro–oesophageal	BCT
Ostrowski et al., 2022, Poland [36]	1	Male, 63, Unspecified	12.9	–	–	LSA diameter 8.3 mm	Aortic arch	Retro-oesophageal	NS
Peña et al., 2013, Columbia [37]	1	Male, unspecified, Columbian	–	–	68	NA	Aortic arch	Retro-oesophageal	NS
Qui et al., 2019, China [40]	5	Female, unspecified, unspecified	8.42	–	–	NA	Aortic arch	Retro-oesophageal	NS
		Male, unspecified, unspecified	5.87	–	–	NA	Aortic arch	Retro-oesophageal	NS
		Male, unspecified, unspecified	10.31	–	–	NA	Aortic arch	Retro-oesophageal	NS
		Male, unspecified, unspecified	9.98	–	–	NA	Aortic arch	Retro-oesophageal	NS
		Male, unspecified, unspecified	1.91	–	–	LSA diameter 1.68	Aortic arch	Retro-oesophageal	BCT
Sakuma et al., 2005,	1	Female, unspecified,	17.2	–	–	LSA diameter	Aortic arch — T4	Retro-oesoph	KD

Japan [41]		unspecified				10.7 mm		ageal	
Sangam et al., 2010, India [42]	1	Male, unspecified, unspecified	12	–	–	LSA diameter 8 mm	Aortic arch (1.4 cm distal to origin of LSA)	Retro-oesophageal	BCT
Suriyonplengsaeng et al., 2014, Thailand [44]	1	Male, 81, Thai	12	–	–	NA	Descending Aorta — T4	Retro-oesophageal	NS

ARSA — aberrant right subclavian artery; BCT — bicarotid trunk; KD — Kommerell’s diverticulum; LSA — left subclavian artery; NA — not available; NS — not specified.