



Incidence and Morphologic Characteristics of Aberrant Subclavian Arteries: A Retrospective Cross-Sectional Study

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Abstract

Aim: The aim of this study to determine the incidence of aberrant right subclavian artery (ARSA) and aberrant left subclavian artery (ALSA), their diameter, angle at their point of origin, distance between them. The cases included in the study were also examined for atrial septal defect (ASD), aneurysm, Kommerell's diverticulum, dysphagia, dyspnea, atherosclerotic heart disease, and hypertension.

Material and Method: This study is a retrospective cross-sectional study conducted at Inonu University Faculty of Medicine Turgut Özal Medical Center. Within the scope of the study, The images of 2365 patients who applied to the Department of Radiology for contrast-enhanced thoracic CT arterial phase imaging were examined. As a result of the review, 52 cases (20 men and 32 women) with ARSA and ALSA were identified and included in the study.

Results: Among the examined images, ARSA was detected in 46 (1.9%) patients, while ALSA was detected in 6 (0.2%) patients. In ARSA cases, ASD and aneurysm were each found in 3 cases. Kommerell's diverticulum was not found in ARSA cases. In ALSA cases, aneurysm was found in 1 case, while Kommerell's diverticulum was found in 5 patients (83.3%). No evidence of ASD was found in ALSA cases. At the origin points, the average diameter of ARSA was 11.7 mm and ALSA was 12.55 mm, with average angles were 76.39° and 60.27°, respectively. The average distance between the right subclavian artery and the left subclavian artery in ARSA cases was 7.27 mm. In ALSA cases, the average distance between the left subclavian artery and the truncus brachiocephalicus was 10.9 mm.

Conclusion: The incidence of ARSA and ALSA in the studied population was 1.9% and 0.2%, respectively. The detailed anatomical characteristics provided in this study can aid in the planning and execution of vascular surgeries involving subclavian arteries.

Keywords: Aberrant subclavian artery; right aortic arch, Kommerell's diverticulum, vascular surgery, anatomical variation

INTRODUCTION

Anomalies and variations of the aortae can be found with many congenital heart diseases (1). In our study, we aimed to draw attention to the importance of these anomalies and variations in terms of prognosis and treatment of diseases.

ARSA is the most common congenital variant of the aortic arch and can occur in 0.4-2% of individuals (2). ARSA arises directly from the aortae, just distal to the left subclavian artery (3). 80% of ARSA cases are retro-esophageal, 15% are between the trachea and esophagus and 5% are anterior to the trachea (3,4).

ALSA; It is a rarer anomaly in society (0.05%-0.1%) (5). ALSA, a branch of the right aorta, is located distal to the right subclavian artery as the first branch of the ascending aorta and usually passes behind the esophagus (6,7).

Kommerell's diverticulum is a rare condition seen together with ARSA and ALSA (5). This diverticulum was named by the German radiologist Burkhard Friedrich Kommerell. Kommerell described this diverticulum as a primitive remnant of the right dorsal aorta. He reported that this anomaly may be a dilatation of the descending aortae (8). ARSA or Kommerell's diverticulum can compress the esophagus or trachea (9). In cases with ARSA, problems occur in the perfusion of the right axillary artery to the right

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carotid artery. Therefore, it is necessary to pay attention to ARSA (9,10).

Congenital anomalies in the aortic arch are caused by branching or position differences. Of these, right-sided aortic arch (RAA) can be seen as three subtypes: RAA with ALSA or rarely RAA with mirror-image branching, RAA with isolated left subclavian artery. These subtypes occur due to different branching of the supra aortic arteries. In isolated left subclavian artery cases, the ALSA is not connected to the arch, but instead to the pulmonary trunk via a patent arterial duct (6).

In our study; we aimed to draw attention to the importance of these anomalies in regional surgical procedures.

MATERIAL AND METHOD

Study Design and Ethical Considerations:

This study is a retrospective cross-sectional study conducted with the permission of İnönü University Health Sciences Non-Interventional Clinical Research Ethics Committee with decision number 2021/2414.

Population and Sample

This study is a retrospective cross-sectional study covering the period January 2015-January 2020 and was conducted at İnönü University Faculty of Medicine Turgut Özal Medical Center.

A total of 25.472 patients who underwent contrast-enhanced thoracic computed tomography (CT) in the Radiology department were identified through a screening of the Hospital Information Management System. Out of these, 2.365 cases were randomly selected for the examination of vascular structures and variations. As a result of arterial phase imaging, 52 cases (20 males and 32 females) with ARSA and ALSA were identified and included in the study.

Imaging Method

The cases were imaged using a new generation 256-slice CT scanner with a stellar detector system (SOMATOM® Definition Flash, Siemens Healthcare, Forchheim, Germany). For each patient, planning images were initially taken for subsequent dynamic imaging positioning. After setting the scan position, 70 ml of iodinated contrast agent (Opaxol 300mg/ml) was administered intravenously through an 18-gauge catheter using an automatic injector at a flow rate of 4-5 ml/sec via the antecubital vein. Post-contrast images were obtained using the following scan parameters: tube voltage 80 kVp, tube current 100 mAs, number of scans 26, scan time 1.5 sec, gantry rotation time 0.28 sec, and detector configuration 128 mm x 0.6 mm. Using the obtained baseline images, reconstructions were performed with slice thicknesses of 2.5 mm and 5 mm.

Evaluation of Images

The data obtained from the imaging were evaluated using the PACS Sectra imaging system (Picture Archiving &

Communications System, Sweden). To facilitate vascular assessment, the images scanned with a slice thickness of 0.625 mm were reconstructed using MIP (Maximum Intensity Projection) to create axial, coronal, and sagittal planes. Angle measurements on the reconstructed images in the PACS system were performed at the level where ARSA and ALSA originate from the aorta, between the aorta and the aberrant vascular structure orifices. The drawings for the angle measurement placed the apex of the angle at the level of the orifice as the vascular structures run parallel to the lumen. Vascular diameter measurements were taken approximately 1 cm distal to the ARSA and ALSA orifices to exclude any narrowing and dilation seen distally.

After the angle and diameter measurements, in cases where ARSA was detected, the distance between the origin of the ARSA and the origin of the left subclavian artery was measured. Similarly, in cases where ALSA was detected, the distance between the origin of the ALSA and the origin of the truncus brachiocephalicus was measured. However, measurement could not be made in a case where the right and left subclavian arteries originated from the same root. Therefore, this case was excluded from the study.

Our study is a retrospective study. Dysphagia, dyspnea, cardiac pathology, vascular aneurysm and atherosclerotic heart disease have been identified as indications. The aim was to evaluate the presence of vascular pathology as a result of the imaging. In addition to measurements, cases with ASD, Kommerell's diverticulum, and aneurysm were identified during the radiological evaluation. Additionally, patient notes, epicrisis report, test results and discharge summaries, from the Hospital Information Management System were evaluated. The number of cases with dysphagia, dyspnea, atherosclerotic heart disease and hypertension were determined from patient files.

To minimize contrast artifacts, the evaluation was performed on the extremity opposite to the one that received the contrast.

Biostatistical Data Analysis

Qualitative data from the variables included in the study were summarized with number (percentage). Compliance of quantitative data with normal distribution was evaluated by Shapiro-Wilk test. Quantitative data that did not show normal distribution were summarized with median (minimum-maximum), while quantitative data that showed normal distribution were summarized with mean \pm standard deviation. In statistical analyses, categorical variables were compared using Fisher's exact chi-square test. For quantitative variables, Mann Whitney U test and Independent sample t test were used for comparisons between two independent groups where appropriate. A value of $p < 0.05$ was considered statistically significant in the applied statistical analyses. All analyses were performed using IBM SPSS Statistics 26.0 for Windows (New York; USA).

RESULTS

The study involved two groups ARSA (n=46) and ALSA (n=6) (Table 1,2). In our study, we made measurements of the diameter, angle and distance of ARSA and ALSA (Table 3). Among the cases, there was one ARSA and

ALSA variation originating from the same root. This case was excluded from the scope of the study because measurements could not be made. There were no other cases in the study in which ARSA and ALSA were seen together.

Variables*		ARSA (n=46)	ALSA (n=6)
Gender [n (%)]	Male	17 (37)	3 (50)
	Female	29 (63)	3 (50)
Atrial septal defect [n (%)]	No	43 (93.5)	6 (100)
	Yes	3 (6.5)	0 (0)
Aneurysm [n (%)]	No	43 (93.5)	5 (83.3)
	Yes	3 (6.5)	1 (16.7)
Kommerell's diverticulum [n (%)]	No	46 (100)	1 (16.7)
	Yes	0 (0)	5 (83.3)

*Variables are expressed in numbers and percentages

Variables		Atrial septal defect		Aneurysm		Kommerell's diverticulum		
		No	Yes	No	Yes	No	Yes	
ARSA (n=46)	Gender [n (%)]	Male	16 (94.12)	1 (5.88)	17 (100.00)	0 (0.00)	17 (100.00)	0 (0.00)
		Female	27 (93.10)	2 (6.90)	26 (89.66)	3 (10.34)	29 (100.00)	0 (0.00)
	p*	1.0		0.286		-		
ALSA (n=6)	Gender [n (%)]	Male	3 (100)	0 (0)	3 (100)	0 (0)	1 (33.33)	2 (66.67)
		Female	3 (100)	0 (0)	2 (66.67)	1 (33.33)	0 (0)	3 (100)
	p*	-		1.0		1.0		

*: Fisher's exact chi-square test

Variables	ARSA (n=46)		ALSA (n=6)		p*
	Mean ± SD	Median (Min-Max)	Mean ± SD	Median (Min-Max)	
Diameter	11.7±3.13	11.3 (4-27)	12.55±2.03	12.7 (10.3-16)	0.289
Angle	76.39±25.49	72.05 (36.2-133)	60.27±11.97	62.95 (45.5-76)	0.197
Distance	7.27±4.24	6.2 (0-23.3)	10.9±10.7	7.25 (3.1-32.4)	0.448

SD: standard deviation, Min: minimum, Max: maximum; *: Mann Whitney U test

Within the scope of the study, cases were also examined in terms of ASD, aneurysm, Kommerell's diverticulum, dysphagia, dyspnea, atherosclerotic heart disease, and hypertension.

ARSA was observed in a total of 46 (1.9%) of 2365 patients included in the study (Figure 1).



Figure 1. Arrow: aberrant right subclavian artery; ARSA is seen coming out of the aortic arc

Considering Table 1; the gender distribution in the ARSA group was 37% male (17 individuals) and 63% female (29 individuals). Within the scope of our study ALSA was observed in 6 (0.2%) of 2365 patients (Figure 2,3). In the ALSA group, the distribution was equal, with 50% male (3 individuals) and 50% female (3 individuals). Regarding the presence of an ASD, 93.5% of the ARSA group (43 individuals) did not have an ASD, while 6.5% (3 individuals) did (Figure 4). In the ALSA group, none of the individuals (100%) had an ASD .

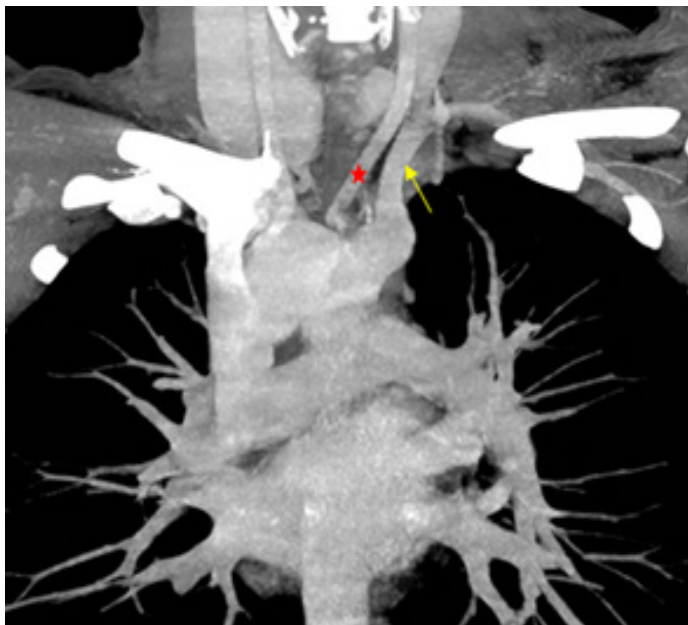


Figure 2. Arrow: Aberrant left subclavian artery; ALSA is seen coming out of the right aortic arc. The left common carotid artery is shown with a red star

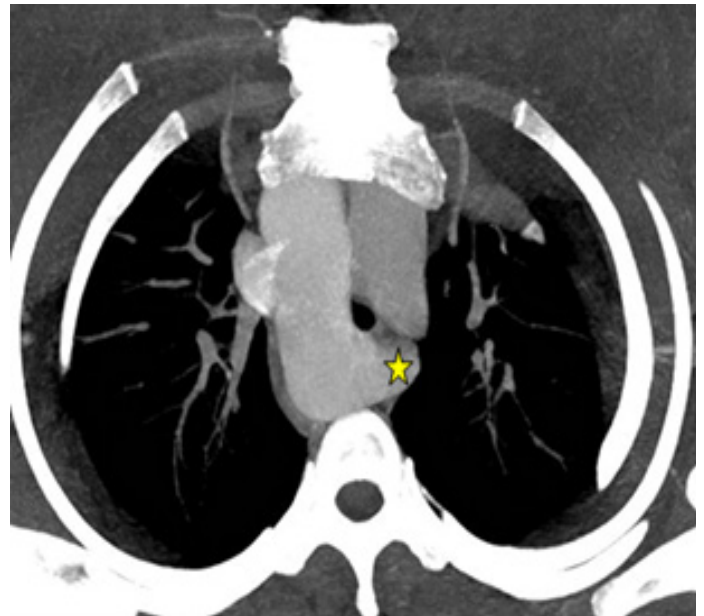


Figure 3. The aberrant left subclavian artery originating from the right aortic arch is indicated by a yellow star

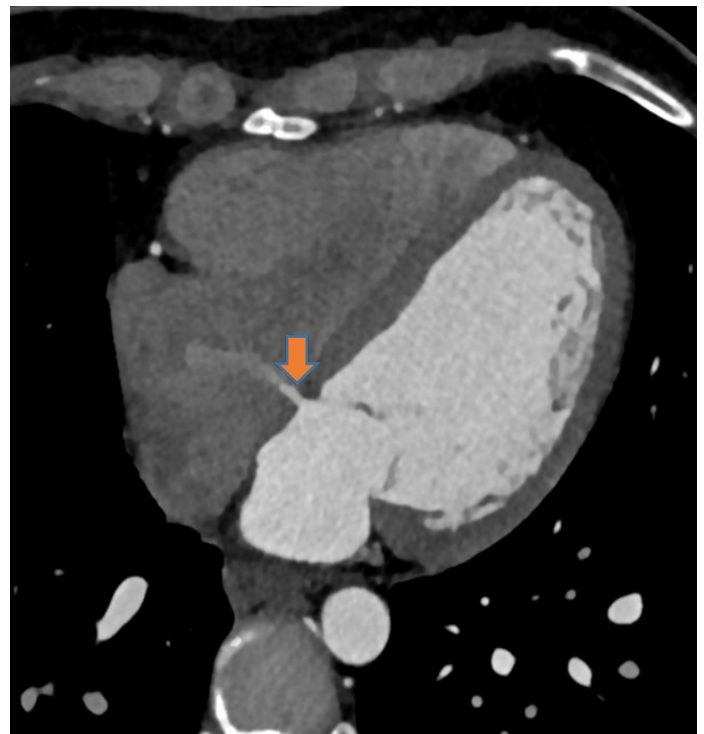


Figure 4. Arrow: Atrial septal defect. In addition to ASD observed at the level of the left and right atrium, passage of contrast material to the right atrium is demonstrated.

When looking at aneurysm occurrence, 93.5% of the ARSA group (43 individuals) had no aneurysm, while 6.5% (3 individuals) had an aneurysm. In the ALSA group, 83.3% (5 individuals) did not have an aneurysm, whereas 16.7% (1 individual) did. For the presence of Kommerell's diverticulum, all individuals in the ARSA group (100%) did not have this condition. In contrast, 16.7% of the ALSA group (1 individual) did not have Kommerell's diverticulum, while 83.3% (5 individuals) did (Figure 5).



Figure 5. Kommerell's diverticulum (★) is seen

The Table 2 presents the distribution of ASD, aneurysm, and Kommerell's diverticulum in ARSA (n=46) and ALSA (n=6) groups, analyzed by gender.

ARSA Group:

ASD: Among males, 94.12% (16 individuals) did not have ASD, while 5.88% (1 individual) did. Among females, 93.10% (27 individuals) were without ASD, while 6.90% (2 individuals) had it. The p-value for gender difference was 1.0, indicating no significant difference.

Aneurysm: All males (100%, 17 individuals) did not have an aneurysm. Among females, 89.66% (26 individuals) were without an aneurysm, while 10.34% (3 individuals) had it. The p-value was 0.286, showing no statistically significant difference between genders.

Kommerell's diverticulum: In the ARSA group, neither males nor females had Kommerell's diverticulum (100%).

We detected the following clinical symptoms in our ARSA cases: Dyspnea 33 (71.7%), hypertension 21 (45.6%), atherosclerotic heart disease 12 (26%), dysphagia lusoria 8 (17.4%).

ALSA Group:

ASD: Both males and females (100%) in the ALSA group did not have ASD.

Aneurysm: Among males, all (100%) did not have an aneurysm. In females, 66.67% (2 individuals) did not have an aneurysm, while 33.33% (1 individual) had it. The p-value was 1.0, indicating no significant gender difference.

Kommerell's diverticulum: In the ALSA group, 33.33% of males (1 individual) did not have Kommerell's diverticulum, while 66.67% (2 individuals) had it. 100% of females (3 individual) had Kommerell's diverticulum. The p-value was 1.0, showing no significant difference between genders.

Other clinical symptoms accompanying ALSA cases were: Dyspnea 3 (50%), hypertension 3 (50%), atherosclerotic heart disease 4 (66.6%). Dysphagia was not observed in the ALSA cases in our study.

The Table 3 shows descriptive statistics for the diameter, angle, and distance of two variation types: ARSA and ALSA.

The mean diameter for ARSA was 11.7 ± 3.13 mm with a median of 11.3 mm (ranging from 4 to 27 mm). For ALSA, the mean diameter was 12.55 ± 2.03 mm with a median of 12.7 mm (ranging from 10.3 to 16 mm). The difference between the two groups was not statistically significant ($p=0.289$). The mean angle for ARSA was 76.39 ± 25.49 degrees, with a median of 72.05 degrees (ranging from 36.2 to 133 degrees). For ALSA, the mean angle was 60.27 ± 11.97 degrees with a median of 62.95 degrees (ranging from 45.5 to 76 degrees). The difference between the two groups was not statistically significant ($p=0.197$).

The mean distance for ARSA was 7.27 ± 4.24 mm, with a median of 6.2 mm (ranging from 0 to 23.3 mm). For ALSA, the mean distance was 10.9 ± 10.7 mm, with a median of 7.25 mm (ranging from 3.1 to 32.4 mm). The difference between the two groups was also not statistically ($p=0.448$).

In all cases of ARSA and ALSA the arteries passed posterior to the esophagus. In none of the cases did the arteries pass between the esophagus and trachea or anterior to the trachea.

DISCUSSION

Within the scope of our study, ARSA was detected in 46 (1.9%) cases and ALSA was detected in 6 (0.2%) cases out of 2365 tomography images. The detected ARSA and ALSA cases were evaluated in terms of ASD, aneurysm, Kommerell's diverticulum, dysphagia, dyspnea, atherosclerotic heart disease, and hypertension. Additionally, diameter, angle and distance measurements were taken in ARSA and ALSA cases.

In a previous study, diameter and angle measurements of the arteria subclavia arteria subclavia were made (11). However, in this study; no information was given about the diameter and angle of ARSA and ALSA. That's why we carried out this study, including ARSA and ALSA measurements.

Additionally, we measured the distances of ARSA cases to the arteria subclavia. In ALSA cases, we measured the distance with the truncus brachiocephalicus. We also evaluated ASD, aneurysm, and Kommerell's diverticulum radiologically. We searched hospital data processing records for dysphagia, dyspnea, atherosclerotic heart disease and hypertension.

Variations of aortic arch diverticula; they are the left aortic arch with ARSA and the RAA with ALSA. The most common congenital anomaly of the aortic arch is the left aortic arch with an ARSA, which occurs in 0.4% to 2.0% of the population (2). Although the incidence of ARSA in our study is slightly higher than the average, it is compatible with the literature. The incidence of ALSA and RAA, which are less common, is 0.05-0.1% (5). The incidence rate we found regarding ALSA is slightly higher than the literature.

Previous study has reported that the co-occurrence of ARSA and aneurysm is rare. Although aneurysm is low in

ARSA, mortality due to rupture (50%) is very high (12,13). In our study, rupture were not observed in ARSA cases. The ARSA case with aneurysm that we found in our study confirms the literature with its low rate.

Although ARSA is asymptomatic in most patients, it causes a type of dysphagia called dysphagia lusoria in 10% of patients due to esophageal compression (14-16). In one study in the literature, the dysphagia rate was stated as 30% (17). The dysphagia rate in our study is compatible with the average of literature studies. The coexistence of ARSA and dyspnea was found to be 23% in the literature. However, the dyspnea rate we found was much higher than the literature. In the same study, it was stated that the rate of coexistence of atherosclerotic heart disease with ARSA was 20-30% (17). The rate of atherosclerotic heart disease in our study is also consistent with the literature. In this study hypertension was found to be (45.6%). However, no study reporting the relationship between ARSA and hypertension has been found in the literature.

In the study of Cina et al. (18), Kommerell's diverticulum was reported in patients with ARSA or ALSA. They have reported that 19% of the asymptomatic Kommerell's diverticulum cases had spontaneous ruptures. Knowing the association of these structures is important to prevent complications. There is literature study showing the association of ARSA with Kommerell's diverticulum at a rate of 15-30% (14). However, in our study, there was no Kommerell's diverticulum or rupture in any of the ARSA cases.

There is literature reporting a 28% association of ARSA with ASD (19). The association of ARSA and ASD we found in our study is quite low.

Studies show that the left subclavian artery connects to the pulmonary artery via the patent ductus arteriosus or ligamentum arteriosum. It has been suggested that it is important to know this variant because it may cause subclavian steal syndrome and vertebrobasilar insufficiency (13).

It was stated that the association of ALSA with RAA and Kommerell's diverticulum is rare (5,20). As an important result in terms of literature, in this study, Kommerell's diverticulum was not seen in any ARSA cases, while it was present in 83% of ALSA cases. Therefore, the relationship of ALSA with Kommerell's diverticulum should be taken into consideration in treatment procedures. Kommerell's diverticulum is thought to be a remnant of the ARSA and ALSA resulting from the developmental anomaly of the fourth dorsal aortic arch during embryological development (21,22). It can be thought that the high prevalence of Kommerell's diverticulum in ALSA cases is due to embryological developmental disorder.

It has been reported in the literature that the association of ALSA and RAA aneurysma is rare (23). The low association of ALSA and aneurysm in our study also confirms the literature.

The association of ALSA with ASD syndrome has been reported in study (24). However, none of the ALSA cases in our study had ASD syndrome (Table 1,2).

In the ALSA cases dysphagia (34%) and dyspnea (25%) has been confirmed in the literature (7). However, in this study ALSA cases, dysphagia was not observed at all, while dyspnea was observed at a rate of 50%. The rate of dyspnea in ALSA cases was found to be twice that of the literature.

In this study, atherosclerotic heart disease (66.6%) and hypertension (50%) were seen with ALSA cases. No study has been found in the literature regarding the clinical findings of atherosclerotic heart disease and hypertension in ALSA cases. We think that the results we found will serve as a resource for future studies.

It has been reported in the literature that isolated left subclavian artery and RAA are rare (0.08%). In the literature, congenital heart diseases associated with isolated left subclavian artery are shown as tetralogy of fallot, right-sided patent ductus arteriosus, large ventricular septal defect, and dextro-transposition of the great arteries (25). None of our cases had isolated left subclavian artery.

The single-center and retrospective study design created a limitation. Multicenter and current studies are needed.

CONCLUSION

Peripheral arterial access is of great importance in ascending and arcus aortic aneurysm surgery and in the surgical treatment of acute aortic dissection. The most commonly used arteries are the right axillary and right subclavian artery. When the lower extremity circulation is stopped by cannulation of these arteries, cerebral perfusion is provided through these arteries. During aortic surgery, the right subclavian artery is cannulated to provide cerebral blood supply. In rare cases, when there is an ARSA, the perfusion given from here will go directly to the descending aortae and will not reach the brain. As a result, brain blood flow will stop. The associated mortality and morbidity rate can be very high. Therefore, this anomaly should be kept in mind while evaluating preoperative tomography images in such cases. And when this anomaly is detected, alternative cannulation sites should be identified and cerebral perfusion should be provided safely. Sometimes the ARSA may be complicated by dissection in the descending region from which the ARSA opens. In these cases, revascularization of the ARSA may be overlooked while treating the dissection of the descending aorta. In this situation, determining the appropriate treatment options is of great importance for the patient to receive a healthy treatment. For this important reasons, we investigated the cases of ARSA.

Our literature review showed that studies investigating variations in the subclavian arteries were not measured the angle, diameter and distance between ARSA and ALSA at the point of exit from the aortic arch. Therefore, we performed the said measurements to provide guidance for the surgical procedures.

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Conflict of interest: The authors have no conflicts of interest to declare.

Ethical approval: This study is a retrospective cross-sectional study conducted with the permission of İnönü University Health Sciences Non-Interventional Clinical Research Ethics Committee with decision number 2021/2414.

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