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MORPHOLOGICAL AND CLINICAL ASPECTS OF ABERRANT SUBCLAVIAN ARTERY: SINGLE CENTER MULTIDEDECTOR COMPUTED TOMOGRAPHY BASED STUDY

Aberran Subklavian Arterin Morfolojik ve Klinik Yönleri: Tek Merkezli Çok Kesitli Bilgisayarlı Tomografi Tabanlı Çalışma

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The study was approved by the local ethics committee with 2020.32.02.06 ethichs number and 02/25/2020 date.

Abstract

Aim: Aberrant subclavian artery (ASA) anomalies are the most common aortic arch malformations. Incidence of these malformations increases with widespread use of computed tomography (CT). Our aim in this study was to investigate the relationship between ASA malformations, gender, age and reasons for admission, and arcus aorta (AA), ASA diameters and AA/ASA diameter ratios.

Materials and Methods: A total 74 patients with ASA were evaluated by thorax CT retrospectively. Patients were divided into aberrant right (ARSA) and left (ALSA) subclavian artery groups. Age, gender, reasons for application, ASA and AA diameters and AA/ASA ratios were evaluated between the groups. Correlations of ASA and AA diameters with age were also evaluated.

Results: 70 of the patients (94.5%) had ARSA, 4 of the patients (5.4%) had ALSA. There was no statistical relationship between gender (p=0.394), age (p=0.443) and reasons for application (p=0.322) between groups. There was no statistical relationship between ASA diameter (p=0.127), AA diameter (p=0.728) and AA/ASA ratio (p=0.339) between groups. There was weak positive correlation with age and diameter of AA (r=0.379, p=0.001), but not with ASA diameter (p=0.059). Moderate positive correlation (r=0.573, p<0.001) was detected between diameters of AA and ASA.

Conclusion: ASA malformations are the most common incidentally detected malformations with increased use of CT. These malformations are not related to age, gender and reasons for application.

Keywords: Vascular malformation, computed tomography, x-ray, aortic arch, subclavian artery.

Öz

Amaç: Aberran subklavian arter(ASA) anomalileri en sık görülen aortik ark malformasyonlarıdır. Bilgisayarlı tomografi (BT) kullanımının artması ile bu malformasyonların görülme sıklığı artmaktadır. Bizim bu çalışmadaki amacımız ASA malformasyonlarının cinsiyet, yaş ve hastaneye başvuru nedenleri arasındaki ilişkiyi araştırmak; arkus aorta (AA), ASA çapları ve AA/ASA çap oranlarının arasındaki ilişkiyi değerlendirmekti.

Materyal ve Metot: ASA'sı olan 74 hastanın toraks BT'leri retrospektif olarak incelendi. Hastalar, aberran sağ subklavian arteri (ARSA) olan ve aberran sol subklavian arteri (ALSA) olanlar şeklinde ikiye ayrıldı. İki grup; yaş, cinsiyet, başvuru nedenleri, ASA ve AA çapları ve AA/ASA oranları açısından karşılaştırıldı. Ayrıca ASA ve AA çaplarının yaş ile korelasyonu değerlendirildi.

Bulgular: Hastaların 70'inde (%94.5) ARSA, 4'ünde (%5.4) ALSA mevcuttu. İki grup arasında cinsiyet (p=0.394), yaş (p=0.443) ve başvuru nedenleri (p=0.322) arasında istatistiksel ilişki bulunmadı. İki grup arasında ASA çapı (p=0.127), AA çapı (p=0.728) ve AA/ASA oranı (p=0.339) arasında istatistiksel ilişki saptanmadı. Yaş ile AA çapı arasında zayıf pozitif korelasyon (r=0.379, p=0.001) izlenirken; yaş ile ASA çapı arasında korelasyon saptanmadı (p=0.059). AA çapı ile ASA çapı arasında orta şiddette pozitif korelasyon (r=0.573, p<0.001) bulundu.

Sonuç: ASA malformasyonları BT kullanımının artmasıyla birlikte daha sık görülen, genellikle insidental olarak saptanan malformasyonlardır. Bu malformasyonların yaş, cinsiyet ve başvuru nedenleri ile ilişkisi bulunmamaktadır.

Anahtar Kelimeler: Vasküler malformasyon, bilgisayarlı tomografi, x-ray, aortik ark, subklavian arter.

INTRODUCTION

Aberrant subclavian artery (ASA) anomalies are the most common aortic arch anomalies. The more common ASA anomaly is aberrant right subclavian artery (ARSA) which has been reported with a

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prevalence of 0.4-2% in the healthy population[1]. Aberrant left subclavian artery (ALSA), which is a more rare ASA anomaly, usually accompanies the right aortic arch (RAA). In the literature, the prevalence of RAA has been reported to be 0.1% in the adult group and is a rare entity[2,3]. Although patients with ASA anomalies are generally asymptomatic, patients may rarely cause dysphagia, dyspnea, cough and recurrent aspirations[4].

Contrast enhanced Computed Tomography (CT) and especially Computed Tomography Angiography (CTA) are important diagnostic methods that replace invasive angiographies in the evaluation of congenital cardiovascular anomalies[5]. The widespread use of CT has led to an ***increase*** in the ***frequency*** of ASA diagnosis. Studies in the literature have focused on ARSA, which is seen more frequently in ASA anomalies. Although there are studies in the litereature that evaluate the incidence of the vascular anomalies such as ARSA and ALSA[6], there is no study in the literature that examines the descriptive properties of these anomalies such as age, gender, and reasons for application.

The aim in this study was to determine the relationships of gender, age and reasons of application of ARSA and ALSA anomalies with contrast-enhanced CT.

MATERIAL METHODS

The study was approved by the local ethics committee with 2020.32.02.06 ethichs number and 02/25/2020 date. Informed consent was obtained from all patients included in the study. In this study, ASA expression was scanned in the hospital PACS system from patients' data who underwent contrast thorax CT between 2012-2019 to investigate different pathologies, and 74 patients with ARSA and ALSA were included to study and evaluated retrospectively.

Patients with ARSA consisted the ARSA group and patients with ALSA consisted the ALSA group, retrospectively. The descriptive statistical relationships of age, gender and reasons for application between ARSA and ALSA groups were evaluated. ARSA and ALSA diameters and aortic arch (AA) diameters were measured in transverse sections of CT images. In addition, the statistical relationship between AA/ASA ratios between ARSA and ALSA groups and the statistical difference of the Kommerell's diverticulum presence in the study groups were evaluated.

CT protocol

Thorax CT images were obtained by Toshiba™ Aquillon™ Prime 80 (Toshiba Medical Systems Corporation, Tokyo Japan) device. CT parameters were assigned as kV: 120, maS: 80, collimation: 1.25 × 1.25 mm, pitch: 1, FOV: 20 × 25 cm, matrix: 512 × 512, and slice thickness: 0.625 mm. Iohexol (Omnipaque 300/100) was used as an intravenous contrast agent at a rate of 4–6 ml/sec. Raw data was processed by an experienced radiologist in the Sectra™ PACS system, and if necessary multi planar reconstruction (MPR) images were obtained.

Statistical Analysis

The distribution of continuous variables is whether normal or not was evaluated by Shapiro-Wilk test. The relationship of gender between the ARSA and ALSA groups was evaluated by Chi-Square test. The relationship of age between the two groups was evaluated by using the Student's t-test. The relationship between gender and ASA diameter was evaluated by Mann Whitney U test. Chi-square test was used to determine the relationship between gender and application reasons. The statistical significance of the presence of Kommerell diverticulum in the ARSA and ALSA groups was evaluated by chi-square test. Student t test was used to determine the relationship of ASA and AA diameters between two groups. Spearman correlation test was used to evaluate the correlation of ASA diameters, AA diameter and AA/ASA ratios with age. Statistical significance was accepted as p <0.05.

RESULTS

41 of the patients were male (55.4%) and 33 of the patients were female (44.6%). 70 (94.5%) of the patients had ARSA (Figure 1) and 4 (5.4%) of patients had ALSA, respectively (Figure 2). The mean age of ARSA patients was 63.3 ± 11.0, and the mean age of ALSA patients was 53.0 ± 23.3. The descriptive statistics of the patients were shown in Table 1. There was no statistically significant relationship of gender and age between ARSA and ALSA groups (Table 1).

Table 1. Descriptivefindings of ASA anomalies

		Vascularmalformation n (%)		Total	*p* value
		ARSA	**ALSA**		
Gender	**Female**	32 (43.2%)	1 (1.3%)	33 (44.5%)	0.394 §
	Male	38 (51.3%)	3 (4%)	41 (55.4%)	
	Total	70 (94.5%)	4 (5.4%)	74 (100%)	
Age	**Female**	64.1±10.4	75		0.443 †
	Male	62.5±11.8	45.6±22.2		

Data were shown as n (%) and mean ± SD.§Chi-square test. † Student t test. ASA: Aberrant subclavian artery, ARSA: Aberrant right subclavian artery, ALSA: Aberrant left subclavian artery, SD: Standart Deviation.

In two patients, the diameter of the ARSA was measured more than 25 mm and was evaluated as compatible with aneurysmatic dilatation.

Kommerell's diverticulum was detected in 13 (18.6%) of ARSA patients and in all of ALSA patients (n = 4, (100%) (Table 2) (Figure 1and 2).

Table 2. The presence of Kommerell'sdiverticulumin thestudygroups

		Kommerell's Diverticulum			*p*value
		Present	**Absent**	**Total**	
Vascular malformation	**ARSA**	13 (18.6%)	57 (81.4%)	70 (100%)	<0.001 §
	ALSA	4 (100%)	0	4 (100%)	
	Total	17 (22.9%)	57 (77%)	74 (100%)	

Data were shown as n (%). § Chi-square test. ARSA: Aberrant right subclavian artery, ALSA: Aberrant left subclavian artery.

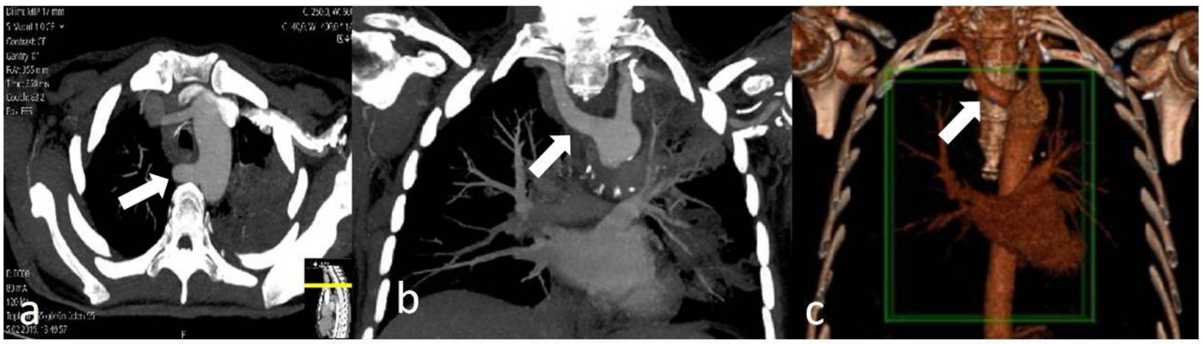

Figure 1. a) Contrast enhanced axial CT scan shows the ARSA (white arrow) which passes behind the eusophagus and trachea. Coronal reformatted (Figure 1b) and VRT (Figure 1c) CT image shows the ARSA (white arrow). ARSA: Aberrant right subclavian artery, CT: Computed Tomography, VRT: Volume Rendered Technic.

Figure 2. a) Axial contrast enhanced thorax CT depicts the RAA with ALSA (white arrow). Coronal refomatted contrast-enhanced CT (Figure 2b) image shows the RAA (black arrow) and ALSA (white arrow) with Kommerell's diverticulum (tailed white arrow). Right subclavian artery (black arrow) can also be seen. VRT (Figure 2c) shows theorigins of great vessels from RAA after removal of thoracal structures. ALSA (white arrow) with Kommerell's diverticulum (white arrowhead), right subclavian artery (tailed white arrow) can be clearly seen. ALSA: Aberrant left subclavian artery, VRT: Volume Rendered Technic, RAA: Right Aortic Arch.

The association of ALSA patients with Kommerell's diverticulum was statistically significant (p <0.001).

There was no statistical relationship between gender and ASA diameter (p = 0.296). There was no statistically significant relationship between patients' application reasons and gender (p = 0.322) (Table 3).

Table 3. Distribution of the cases according to the reasons for application.

		VascularMalformation		***p*value**
		ARSA n(%)	**ALSA n(%)**	
Reason for application	Pneumothorax	3 (4.2%)	-	0.322 §
	COPD	4 (5.7%)	1 (25%)	
	Chest pain	6 (8.5%)	-	
	Cough	14 (20%)	1 (25%)	
	Malignancy	28 (40%)	-	
	Chest Trauma	3 (4.2%)	-	
	PPD	5 (7.1%)	-	
	Cerebrovascular Accent	4 (5.7%)	-	
	Pulmonary embolus	3 (4.2%)	2 (50%)	
	Total	70 (100%)	4 (100%)	

Data were shown as n(%). § Chi-Square test.ALSA: Aberrant left subclavian artery, ARSA: Aberrant right subclavian artery, COPD: Chronic obstructive pulmonary disease, PPD: Pulmonary paranchimal disorders

There was no statistically significant difference in ASA diameters (p = 0.127), AA diameters (p = 0.728) and AA / ASA ratios (p = 0.339) between the two groups (Table 4).

Table 4. Statistical evaluation of ASA, AA diametersand AA / ASA diameterratios in the ARSA and ALSA groups.

	ARSA	**ALSA**	***p*value**
ASA diameter	12.01±4.28	9.50±1.91	0.127 †
AA diameter	28.04±4.76	26.75±6.13	0.728 †
AA/ASA ratio	2.46±0.51	2.91±0.95	0.339 †

Data were shown as mean±SD. † Mann Whitney U test. ASA: Aberrant subclavian artery. AA: Arcus aorta. ARSA: Aberrant right subclavian artery. ALSA: Aberrant left subclavian artery. SD: Standart Deviation**.**

A weak positive correlation was found between age and AA diameter (r = 0.379, p = 0.001), and a moderate positive correlation between ASA diameter and AA diameter (r = 0.573, p <0.001). No significant correlation was observed between age and ASA diameter (p = 0.059)

DISCUSSION

Our study showed that diameters of ARSA and ALSA, which are most seen among the aortic arch anomalies, do not depend on gender and reasons of applications, and do not increase with age. In our study, the reasons for patients admitted to the hospital were generally due to reasons other than the causes of ASA anomalies and were incidental. In addition, our study predicted that ASA anomalies can be seen more frequently incidentally with increasing use of CT.

There are six branchial archs that begin in the second week and develop in the seventh week, which extends from cranial to caudal in embryological life. These structures consist of vascular archs which are seen at different time periods that develop as right and left pairs and merge with the ventral and dorsal aorta. The first, second and fifth vascular archs do not develop and undergo involution. The third, fourth and sixth branchial branches develop the carotid, subclavian system, aortic arch and pulmonary artery systems. The arch of the aorta develops from the fourth arch, and the pulmonary artery systems and truncus arteriosus develops from the sixth archs, respectively[7,8].

ARSA is the most common anomaly developing from the left aortic arch. ARSA can be seen when there is involution between the right main carotid and right subclavian arteries during the involution of the right aortic arch, and the right subclvian artery feeds the right upper extremity as the last branch of the left aortic arch. It has been reported that 80% of the cases pass through the posterior esophagus, 15% between the trachea and esophagus, and 5 % between the main bronchi[9]. Dysphagia due to esophageal compression is the most common symptom (dysphagia lusoria). Horner syndrome due to nerve compression and cough has also been reported[10]. ARSA was first described by Bayford in 1794 during the autopsy of a 62-year-old woman who died of dysphagia. ARSA was shown to compress the esophagus in this autopsy[11].

RAA has been reported as approximately 0.04% to 0.1% in the series of necropsy[12]. However, the incidence determined by CT is estimated to be higher. RAA often coexists with anomalies such as cardiac anomalies, tracheoesophageal fistula, and esophageal atresia[13].

Three types of RAA are defined:

a) Type I: Mirror-type branching

b) Type II: RAA with ALSA

c) Type III: RAA associated with Isolated Left subclavian artery

The most common of these types is type II, which is associated with ALSA. ALSA anomalies are observed less frequently than ARSA, and they are more frequently associated with RAA[14]. Although the prevalence of ALSA is observed less than ARSA, cardiac anomalies are more common[12]. In their study, Murakoa et al. reported that ALSA stenosis can be seen together with ALSA / RAA complex[2].

Kommerell's divertuculum was defined as a developmental error accompanied by the remnant of the fourth dorsal aortic arch[15]. Kommerell's diverticulum was first reported and described by German radiologist Dr. Buckhard F. Kommerell in 1936[16]. Kommerell's diverticulum is defined as the protrusion of the proximal aorta, which can be associated with the right and left aortic archs, where the subclavian artery root originates. Kommerell's diverticulum can accompany both ARSA and ALSA anomalies. Kommerell's diverticulum accompanying ARSA anomalies tend to be wide and round, while Kommerell's diverticulum associated with ALSA are generally observed as cone shapes[7].

In our study, Kommerell's diverticulum was observed in all ALSA patients and in 18.6% of ARSA patients. The findings in our study for Kommerell's diverticulum were consistent with the prevalence values specified in the literature for ARSA patients. On the other hand, since the number of ALSA patients was not sufficient in our study, the association of Kommerell's diverticulum with ALSA was not compatible with the literature. Therefore, studies with higher patient numbers are needed[14]. Among the Kommerell's divertuculumes in our study, those associated with ARSA were diverticular formations in the form of pouch in the near the origin of ARSA. The Kommerell's diverticulumes associated with ALSA were the cone-shaped diverticula, as described in the literature (ALSA).

Our study has shown that ASA diameter increases with AA correlation with increasing age. However, the findings in our study showed that, except for two ARSA aneurysm patients, this raise was increases in correlation with AA diameter. But ASA anomalies do not cause hemodynamic changes that may cause abnormal diameter increase when compared with aorta. This is significant in terms of showing why this patient group is not presented with hemodynamic disorders.

In the literature, asymptomatic presentation of ASA anomalies has been reported. Besides, there are also studies related to the fact that ARSA cases are most frequently presented with dysphagia in terms of vascular compression[17]. In addition, in the pediatric population, cases with chronic cough and shortness of breath have been reported[18]. In our study, no relationship was observed between the reasons for admission to the hospital and the occurrence of ASA because of ASA anomalies are usually presented as symptomatically. Another reason is that ASA cases are seen more frequently incidentally due to the increased frequency of using CT [19].

The limitations of our study were the retrospective study design and the number of patients being less prominent in the ALSA group.

CONCLUSION

ASA anomalies are anomalies which do not disrupt hemodynamics and are usually asymptomatic. These patients generally admit to the hospital for reasons other than vascular compression and hemodynamic reasons that might be caused by ARSA or ALSA and are these anomalies are generally diagnosed incidentally. With widespread use of CT, ASA anomalies are seen more frequently and incidentally.

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