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Evaluation of Congenital Mediastinal Vascular Anomaly Types and Frequencies Among 2000 Cases.

2000 Hastada Konjenital Mediastinal Vasküler Anomali Tiplerinin ve Sıklığının Değerlendirilmesi

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ABSTRACT

Aim: In this study, it was aimed to examine the incidence of congenital thoracic vascular anomalies in our region.
Methods: The features of the patients with non-specific complaints who underwent a thorax Computed Tomography over a two years period were retrospectively reviewed on the hospital database, and demographical data (Gender and age) were recorded. All the Computed Tomography images were interpreted regarding thoracic vascular anomalies, including persistent left superior vena cava, azygos lobe, aberrant right subclavian artery, dilated left superior intercostal vein, right-sided aortic arch, situs inversus, and partial anomalous pulmonary venous return. The incidences of these thoracic vascular anomalies were calculated and compared with previous studies.
Results: Mediastinal vascular anomaly was detected in 62 (3.1%) patients. A mediastinal vascular anomaly was observed in 27 (3.5%) female patients and 35 (2.6%) male patients. The most common mediastinal vascular anomaly in this study was the right aberrant subclavian artery (n:17, 0.9%) and the rarest was partial anomalous pulmonary venous return (n:1, 0.1%). Persistent left superior vena cava incidence was 0.3% (n:6), azygos lobe 0.7% (n:14), right-sided aortic arch 0.3% (n:5), situs inversus totalis 0.2% (n:3), and dilated left superior intercostal vein 0.8% (n:16).
Conclusion: Mediastinal vascular anomalies are rare and usually asymptomatic. But the imaging findings of these conditions must be well-known to accurately planning the interventions and also to prevent iatrogenic injuries.

Key Words: Computed Tomography, Partial anomalous pulmonary venous return, Aberrant right subclavian artery, Situs inversus totalis, Dilated left superior intercostal vein

ÖZ

Amaç: Bu çalışmamızda bölgemizde konjenital mediastinal vasküler anomalilerin sıklığını araştırmayı planladık.
Yöntem: Yaklaşık 2 yıllık bir süreçte non-spesifik semptomlar ile kontrastlı toraks Bilgisayarlı Tomografi çekilen hastaların özellikleri hastane veri tabanından retrospektif olarak tarandı. Yaş ve cinsiyet gibi demografik özellikler kaydedildi. Tüm Bilgisayarlı Tomografi görüntüleri, persistan sol superior vena kava, azygos lobu, aberran sağ subklavien arter, dilate sol superior interkostal ven, sağ taraflı arkus aorta, situs inversus ve parsiyel anormal pulmoner venöz dönüş anomalileri açısından yorumlandı. Çalışmamızda bulunan insidans değerleri ile önceki çalışmaların sonuçları karşılaştırıldı.
Bulgular: Mediastinal vasküler anomali 62 (%3.1) hastada saptandı. Kadın hastaların 27 (%3.5)'inde ve erkek hastaların 35 (%2.6) 'inde bir mediastinal vasküler anomali gözlemlendi. Çalışmamızdaki en sık saptanan mediastinal vasküler anomali, sağ aberran subklavien arter (n:17, %0.9) ve en nadir görüleni ise parsiyel anormal pulmoner venöz dönüş anomalisi idi (n:1, %0.1). Persistan sol superior vena kava insidansı %0.3 (n:6), azygos lobu %0.7 (n:14), sağ taraflı arkus aorta %0.3 (n:5), situs inversus totalis %0.2 (n:3) ve genişlemiş sol superior interkostal ven ise %0.8 (n:16) olarak bulundu.
Sonuç: Mediastinal vasküler anomaliler nadir olup sıklıkla asemptomatiktir. Ancak bu durumların görüntüleme bulgularının iyi bilinmesi, girişimsel işlemleri doğru biçimde planlamada ve aynı zamanda iatrojenik yaralanmaları önlemede gereklidir.

Anahtar Kelimeler: Bilgisayarlı Tomografi, Parsiyel anormal pulmoner venöz dönüş anomalisi, Sağ aberran subklavien arter, Situs inversus totalis, Genişlemiş sol superior interkostal ven

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INTRODUCTION

Various development conditions may affect the anatomy of intrathoracic arterial and venous structures including superior vena cava, pulmonary veins, aorta and their branches. The variations of these vascular structures may be isolated or associated with a syndrome. Most of these anomalies are asymptomatic and are detected incidentally on thorax imaging. But for especially accurate pre-operative/intervention evaluation of the thoracic vascular structures, the vascular anatomical variations of the thorax must be well-known. Computed Tomography (CT) is the most frequently performed imaging modality for detection of these anomalies while Magnetic Resonance Angiography and conventional catheter angiography can be also used. CT has excellent spatial resolution but a high-radiation dose is its' principal disadvantage. For diagnosis of thoracic vascular anomalies, conventional catheter angiography is accepted as the gold standard method, but it is an invasive procedure that involves ionizing radiation. Magnetic Resonance Imaging (MRI) is a non-invasive modality not involving ionizing radiation although it has a poor spatial resolution, when compared with CT and conventional catheter angiography [1].

In this study, we aimed to evaluate the incidence of congenital mediastinal vascular anomalies among patients with non-specific complaints in our region, and to compare these results with previous studies.

MATERIAL AND METHODS

Patients

The patients who underwent a CT of the thorax in a tertiary chest disease hospital with non-specific complaints including cough, dyspnea and chest pain, between the years 2011 and 2013, were included in this study. Demographical-medical data and the CT images of the patients were reviewed on the hospital database. The patients with a previous thoracic-vascular surgery, arterial and venous thrombosis or systemic, or thoracic chronic disease history were excluded from this study. Because we aimed to evaluate congenital Mediastinal vascular anomalies.

This retrospective study was approved by the institutional ethics committee (Ankara Atatürk Chest Diseases and Chest Surgery Training and Educational Hospital ethics committee) with a number of 005070 on 22.08.2013. No written informed consent could be obtained from the patients because of the retrospective nature of the study.

CT technique

A Somatom Emotion 6 scanner system (Siemens Medical Systems, Forchheim, Germany), with a pitch of 0.75, a 6x3 mm collimation and a reconstruction thickness of 10 mm were used to obtain CT images. In all patients, an intravenous iodine contrast agent (iohexol-300) was administrated with a dose of 1.2 ml/kg at speed of 3-4 mL/s. Images were obtained from common carotid artery bifurcation level to the level of renal artery origins and the image withdrawal was started 25th second after contrast agent administration. A cut-off diameter >4mm was considered as significant for the definition of a dilated left superior intercostal vein. Images were interpreted by two radiologists with 14 and 11 years of thoracic imaging experience in consensus regarding thoracic vascular anomalies mentioned above.

Statistical Analysis

The descriptive variables were presented as the mean ± standard deviation, minimum/maximum, and percent (%). For the statistical analysis, a software program (IBM SPSS Statistics for Windows, Version 21.0. Armonk, NY: IBM Corp., USA) was used.

RESULTS

Nineteen patients with previous thoracic surgery, 43 with chronic diseases and 36 whose medical data was not available were excluded from the study. A total of 2 000 patients (mean age 56.13±14.62, 12-93 years) were included in this study. Among them 776 were female (38.8%) (mean age 56.93±15.19, 13-93) and 1 224 were male (61.2%) (mean age 55.63±14.23, 12-88 years). Female/male ratio was 0.63. A thoracic vascular anomaly was detected in 3.1% (n:62) of the patients while in 96.9% (n:1938) of them

no anomaly was observed. A thoracic vascular anomaly was observed in 3.5% (27/776) of the females and 2.6% (25/1224) of the males. The detailed features of the patients are summarized in Table 1.

Table 1: Prevalence of mediastinal vascular anomalies among males and females

Variation	Female		Male		Total	
	N	%	N	%	N	%
No anomaly	749	96.5	1189	97.1	1938	96.9
Aberrant right subclavian artery	10	1.3	7	0,6	17	0.9
Dilated left superior intercostal vein	7	0.9	9	0.7	16	0.8
Azygos lobe	3	0.4	11	0,9	14	0.7
Persistent left superior vena cava	2	0.3	4	0,3	6	0,3
Right-sided aortic arch	4	0.5	1	0.1	5	0.3
Situs inversus	1	0.1	2	0.2	3	0.2
PAPVR*	0	0.0	1	0.1	1	0.1
Total	776	38.8	1224	61.2	2000	100

*PAPVR: Partial anomalous pulmonary venous return

The most common vascular variation in this study was aberrant right subclavian artery which was detected in 17 (0.9%) patients. Dilated left superior intercostal vein (DLSIV) (Figure 1) was observed in 16 (0.8%), azygos lobe in 14 (0.7%), persistent left superior vena cava (PLSVC) (Figure 2) in 6 (0.3%), right-sided arcus aorta (Figure 3) in 5 (0.3%), and situs inversus in 3 (0.2%) patients. Partial anomalous pulmonary venous return (PAPVR) (Figure 4) was the rarest thoracic vascular variation, which was seen in one (0.1%) patient. Normal venous anatomy is illustrated in Figure 5.

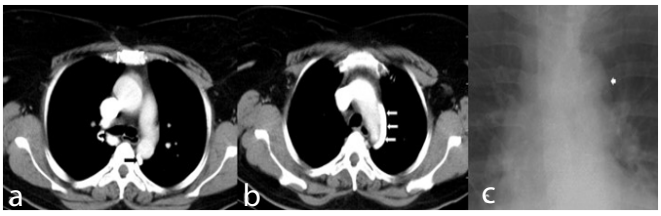

Figure 1a-c. (Dilated left superior intercostal vein) Dilatation of both azygos vein (curved arrow), accessory hemiazygos vein (black arrow) and left superior intercostal vein (white arrows) in a patient with congestive heart failure on axial CT images (a,b). The "aortic nipple" sign (little white arrow) is seen close to aortic knob on chest x-ray (c).

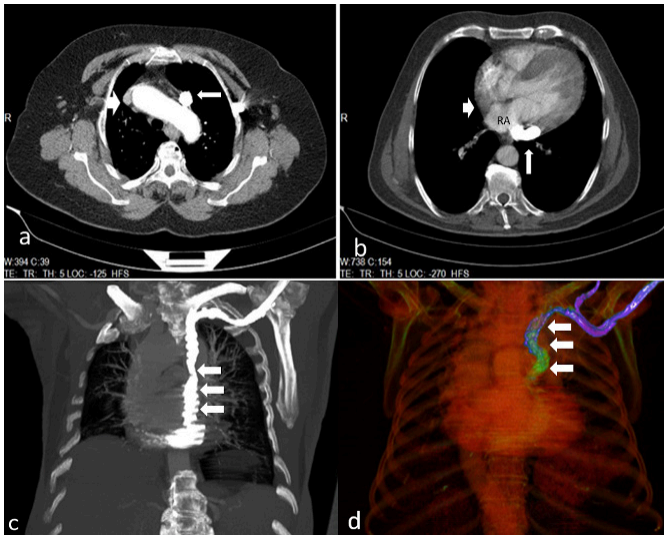

Figure 2a-d. (Persistent left superior vena cava) Right superior vena cava (short arrows) left superior vena cava (long arrows) that draining to right atrium (RA).

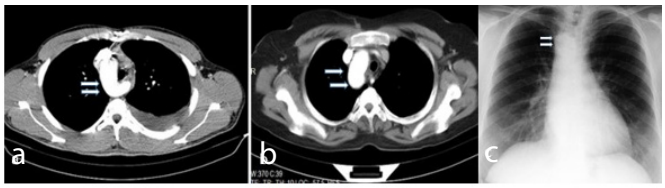

Figure 3a-c. (Right-sided aortic arch) Arcus aorta coursing to the right of the esophagus and trachea (white arrows) on axial CT images (a,b) and on chest x-ray (c).

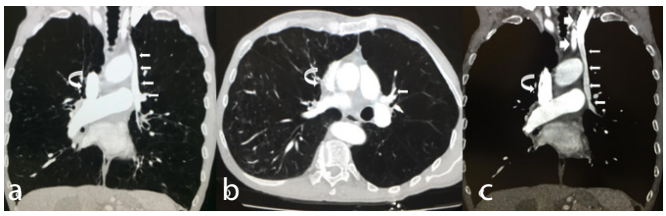

Figure 4a-c. (Partial anomalous pulmonary venous return) Left superior pulmonary vein (Long arrows) joins to left brachiocephalic vein (short arrows) and drained to superior vena cava (curved arrow) and then to right atrium.

DISCUSSION

Persistent left superior vena cava (PLSVC) is the most frequent congenital venous anomaly of the thorax that is reported in 0.5% of the general population and 4% of the patients with congenital cardiac diseases [2,3]. PLSVC occurs due to failure in regression of the left anterior cardinal vein and is frequently accompanied by right-sided superior vena cava. However, it may be rarely detected in isolation when the right anterior cardinal vein regressed. If a superior vena cava

duplication is present, left superior vena cava lies caudally lateral to mediastinum and anterior to the left hilus [4].

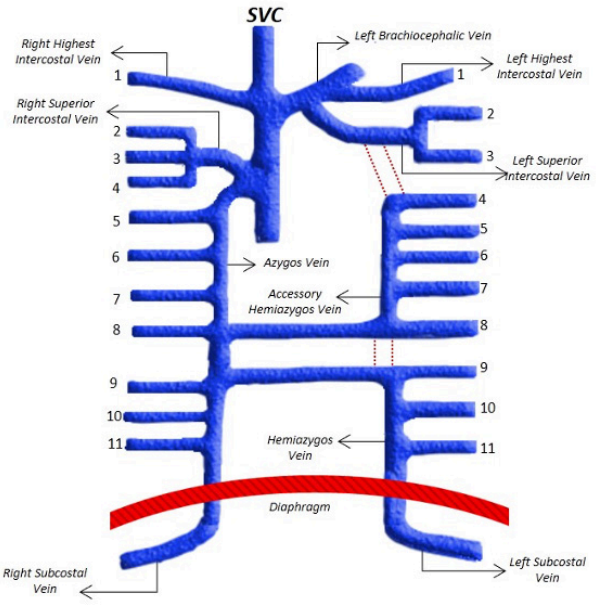

Figure 5. (Normal venous anatomy of mediastinum) The illustration of venous structures of the mediastinum.

PLSVC is a usually asymptomatic condition that is detected incidentally on thorax CT or x-ray imaging. Left or right cardiac enlargement in the patients with PLSVC must raise a suspicion of an atrial or ventricular septal defect. It has no clinical importance even in the patients with right-to-left shunt also when drained to left atrium. But when detected, it must be described for optimal planning of implantation of cardiac pacemakers or venous ports [5-7]. The incidence of the condition was suggested to be between 0.3-0.5% in recent studies [5,6]. We detected PLSVC in 0.3% (n:6) of our patients. Among these 6 patients, two (0.3%) were female and the remaining four (0.3%) were male. In all these cases right superior vena cava was also present. Our results of PLSVC were consistent with previous studies and the incidence of the condition in males and females is the same.

An anomalous course of the azygos vein in apex of the right lung results in azygos lobe that is detected in 0.42-1.2% of the general population [8]. In most cases, the diagnosis can be made by x-ray while in some patients CT may be required [9]. The incidence of the condition was reported as 0.4% on x-ray and 1.2% on CT images with a male predominance [10]. In this study, we observed azygos lobe in 0.7% (n:14) of the patients. Consistent with the literature, the incidence of the condition in males (0.9%, n:11) was markedly higher than in females (0.4%, n:3). Azygos lobe is almost always asymptomatic and does not require intervention. Azygos lobe may be misdiagnosed as an abscess or air cyst on chest x-ray. Also, consolidation in the azygos lobe may be misinterpreted as a malignant tumor [11].

The most frequent congenital anomaly of aortic arch is the aberrant right subclavian artery, which is seen in 0.5-2% of the general population [12]. The condition occurs from the regression of right fourth innominate artery, the persistence of the seventh intersegmental artery, and the abnormal arise of the right subclavian artery from the aortic arch as the last branch in embryonic period [13]. It crosses upwards and to the right in the mediastinum. The condition is usually asymptomatic unless it compresses by the esophagus or trachea [12]. Computed Tomography is more sensitive (100%) for the recognition of aberrant subclavian artery than x-ray (92%) and doppler ultrasound (20%) [14]. Previous studies suggested the incidence of aberrant right subclavian artery as 0.4%-0.5% [15,16]. In our study, we detected this condition in 0.9% (n:17) of the patients. When compared with previous studies we detected a higher incidence. Among 17 patients with aberrant right subclavian artery, ten were female and seven were male. In the literature, the condition was more commonly observed in females than males (58% versus 42%, respectively) [17]. In our study, consistent with the literature, we also found female dominance.

The right-sided aortic arch occurs by the total obliteration of the left fourth aortic arch and left dorsal aorta and the development of the right fourth aortic arch and right dorsal aorta. The major subtypes of the condition are: 1-with mirror image branching, 2-with aberrant left brachiocephalic artery 3-with aberrant left subclavian artery, 4-with isolated left subclavian artery. Aortic arch anomalies may be detected by x-ray but usually CT and MRI are required for the confirmation [18,19]. The right-sided aortic arch is detected in 0.1% of the adults [19]. In this study, the incidence of the condition was 0.3% (n:5). Among the five patients, four (0.5%) were female and the remaining one (0.1%) was male.

Partial anomalous pulmonary venous return (PAPVR) is another congenital vascular anomaly that is characterized by the drainage of one or more pulmonary veins to systemic circulation or right atrium [20,21]. Four variants of PAPVR have been reported: supra-cardiac, cardiac, infra-cardiac and mixed [22]. The most common venous return anomaly is the drainage of the right superior pulmonary vein to superior vena cava or right atrium. In some cases, the right pulmonary vein may drain to inferior vena cava (Scimitar syndrome). Very rarely, drainage to azygos-portal and hepatic veins is reported. Left pulmonary veins drain to left brachiocephalic vein or coronary sinus [23-24]. The only case of PAPVR in our study was a supracardiac type in which the left superior pulmonary vein joins the left brachiocephalic vein and then drains to the right atrium.

PAPVR is rare, as detected in an incidence of 0.4% in pediatric autopsy series [23]. Ho et al, reviewed thorax CT images of 45 538 individuals and detected PAPVR in 0.1% of them. In their series, 58% of the individuals were female and 42% were male [20]. In our study, the incidence of the condition was found to be 0.05%, as the only patient with PAPVR was male.

PAPVR is usually asymptomatic as does not require treatment unless accompanied by other severe cardiovascular pathologies. But awareness of this condition is important to prevent iatrogenic complications such as malposition, perforation, or thrombosis, during and after interventions [25].

Situs inversus totalis is characterized by the reversion of the major visceral organs from their normal locations. In this condition, not only the heart and bronchial tree but also the intestines and mesenteric structures are mirrored from their normal position. Jejunum, stomach, descending colon are seen on the right side while ileum, ascending colon and the Treitz ligament are located in left side. It is seen in approximately 1/8 000-25 000 and the diagnosis is usually made in childhood based on radiological examinations. Sometimes congenital heart defects and immune deficiency syndromes are associated with situs inversus. In isolated cases, the condition is usually detected in adulthood when being evaluated for non-specific complaints [26]. In our study, two males (0.2%) and one female (0.1%) with situs inversus were identified. Total situs inversus incidence in the study was 0.2%.

The earlier diagnosis of situs inversus is essential for the accurate interpretation of the visceral organ complaints which are not localized to their normal position, as well as to prevent iatrogenic injuries in the interventions.

Intercostal veins drain into the hemiazygos, whereas accessory hemiazygos veins drain into the left side and into the azygos vein in right. Right superior intercostal vein joins to azygos vein above right main bronchus and close to the proximal segment of aortic arch. Left superior intercostal vein forms by union of 2nd and 3rd (and sometimes 4th) intercostal veins. In 75% of the individuals, the left intercostal vein joins to accessory hemiazygos vein and then drains to the left brachiocephalic vein. It is seen close to the aortic knob on chest x-rays and is termed as ‘aortic nipple’. It is seen in the chest x-rays of 1.4-9.5% of healthy individuals [27]. When the diameter of this vessel is >4 mm, it is termed as Dilated Left Superior Intercostal Vein (DLSIV). The patients with DLSIV must be further examined regarding venous anomalies. Inferior-superior vena cava occlusions, congestive heart failure, Budd-Chiari syndrome, left brachiocephalic vein hypoplasia, accessory hemiazygos vein hypoplasia and azygos vein aplasia, may be associated with DLSIV [28]. In this study, DLSIV was detected in 0.8% (n:16) of the patients and among these, nine were male and seven were female. Of 16 patients with dilated left superior intercostal vein, three had hypoplasia of hemiazygos vein and two had congestive heart failure. No additional findings were observed in the remained eleven patients on imaging.

Limitations: The limitations of this study are that it is a retrospective and single-center study.

Conclusion

Mediastinal vascular anomalies are usually asymptomatic and detected incidentally on the imaging for various complaints. However, these anomalies must be well identified for an accurate diagnosis and optimal planning before intervention, in order to prevent iatrogenic

complications. To evaluate the prior imaging of the patients undergoing interventional procedures is essential to investigate mediastinal vascular anomalies. In addition, a preliminary MRI or CT angiography may be performed to detect possible associated vascular anomalies in the patients with congenital heart defects or suspicious clinical (such as collaterals) and radiological (such as mediastinal widening on chest X-rays) findings.

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Ethics Committee Approval: National and international ethical rules were observed in this study. Ethic Board: Atatürk Chest Diseases and Thoracic Surgery Training and Research Hospital, Board name, date and number of 005070 on 22.08.2013.

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