Case Report

A Rare Aortic Arch Anomaly: Combination of Vertebral Arteria Lusoria with Kommerell's Diverticulum, Bovine Aortic Arch, and Left Vertebral Artery with Extreme Proximal Origin

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Abstract

Vertebral arteria lusoria (VAL), defined as the right vertebral artery (VA) originating from the aortic arch distally to the left subclavian artery (SCA) and progressing retroesophageally, is a rare vascular anomaly. This anomaly is rarely associated with abnormalities such as aortic coarctation, Bovine aortic arch, and Kommerell's diverticulum. It is uncommon for VAs to originate from the SCA below the level of the 1st rib. To our knowledge, no instance of VAL has been described, in which the left VA originates from the SCA below the first rib. We aimed to present computed tomography findings in a patient with VAL, Kommerell's diverticulum, Bovine aortic arch, and a left VA with the extreme proximal origin, which could be also defined as "vertebrosubclavian trunk," in this case report.

Keywords: *Bovine aortic arch, Kommerell's diverticulum, vertebral arteria lusoria, vertebral artery with extreme proximal origin*

Introduction

The "typical" three-branch aortic arch pattern includes: the brachiocephalic trunk that gives off the right subclavian artery (SCA) and the right common carotid artery (CCA), the left CCA, and the left SCA.[1]

Vertebral artery (VA) typically originates (50.6%–99.9%) from the posterosuperior aspect of the 1st part of the SCA, 0.5–2 cm medial to the thyrocervical trunk origin.[1] Aberrant origin of the VAs is not very usual, with a reported incidence of 4.7% in a recent systematic literature review based on 32 original studies encompassing 14738 subjects.[1] Variations of the VA origin usually occur on the left side, and rarely occur on the right side with a reported incidence of only 0.69%.[1,2] The most common reported atypical VA origin is from the aortic arch with an incidence of 5.5%. A left VA with an aortic arch origin is the commonest reported variation (range 0.79%–8%) most frequently located in between left CCA and left SCA, while most rarely originates distal to left SCA.[1] The aortic origin of the right VA and especially its origin as the last branch is very rare.[1,2]

In rare cases, the right VA originates from the aorta, and even more rarely from the descending part. Like the classic arteria lusoria, it usually takes a retroesophageal course, probably the main reason why this VA variant is also named lusoria. Like the classic arteria lusoria, the vertebral arteria lusoria (VAL) has been shown to originate from Kommerell's diverticulum.[2] Bilateral aortic origin of the VA has been reported, too.[1] The VA origin below the level of the first rib has been reported in the literature as a rare entity with an incidence of 2.94%, without definition of right or left direction (3 out of 102 cases). Such condition, which is the extreme proximal origin of left VA, has been also reported as "vertebrosubclavian trunk" in the literature with an incidence of 0.3%–0.73%.[3-5]

In cross-sectional radiological imaging, vascular anatomical variations of the aortic arch are often encountered incidentally.[6] These variations of great vessels are generally asymptomatic and can be noticed later in life. However, it may rarely be closely related to tracheoesophageal structures and cause symptoms due to variation of the course.[2,6] We aimed to present computed tomography (CT) findings of a rarely seen VAL abnormality accompanied by

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Kommerell's diverticulum, bovine aortic arch, and a left VA with the extreme proximal origin, which could be also defined as "vertebrosubclavian trunk," in this case report.

Case Report

An 82-year-old male patient, who had no complaints and was followed up for a history of lymphoma, underwent thorax CT for follow-up. Incidentally, it was observed that the right VA originated from the aortic arch distal to the left SCA and had a retroesophageal course. Focal dilatation consistent with Kommerell's diverticulum was observed in the origin of the right VA [Figure 1a and b]. The innominate artery (IA) and left CCA emerged as a single trunk from the aortic arch, which is consistent with the Bovine aortic arch. Furthermore, the left VA, as the first branch of the left SCA, originated from the more proximal and posteromedial part of the left SCA [Figure 2a and b]. The patient did not have any symptoms related to these variations.

Discussion

The most common variation in the aortic arch branching pattern is the so-called "Bovine arch" in which the IA and left CCA fuse as a single root.[6] This variation was also observed in our case. Normally, VAs arise as the first branch from the posterosuperior parts of the ipsilateral SCAs and above the level of the 1st rib (97.1%).[6,7] Many papers have been published about the types of anomalies of origin of the vertebral arteries in the literature. The most common VA variation is the type, in which the left VA arises directly from the aortic arch in the region between the left CCA and the left SCA and accounts for 2.4%–5.8% of instances.[7] In our case, the left VA emerged as the first branch of the left SCA as expected, but its origin was caudal to the 1st rib level. The VA origin below the level of the first rib may be encountered rarely, and its incidence has been reported as 2.94%, without definition of right or left direction (3 out of 102 cases).[7] The proximal origin of left VA from the left SCA could be also referred as "vertebrosubclavian trunk" and its incidence in a radiological study was found to be 0.3% (8 out of 2287 cases), and 0.73% in another study in the literature.[3-5]

Variations in the right VA are seen less frequently, and the most common type is that the right VA emerges from the root of the right SCA (76.6%).[7] In our case, the right VA originated as the last branch of the aorta and had a retroesophageal course. Therefore, this anomaly is called VAL.[2,8] The incidence of VAL has been reported very low in the literature.[2,7,9,10] Its association with anomalies such as coarctation of the aorta, Bovine arch, and bilateral VAs originating from the aortic arch have been published in a limited number of articles.[6,9-11] As in our case, its association with focal enlargement at the level of origin called Kommerell's diverticulum is also rare.[2,8,9,12] To our knowledge, extreme proximal left VA origin from the left SCA, caudal to the 1st rib, has not been defined before together with these anomalies.

In 1948, Edwards first proposed the double aortic arch system hypothesis to explain the embryological process of branching.[13] According to this hypothesis, most congenital aortic arch anomalies are caused by the persistence of a segment that should normally regress or regression of a segment that should persist. VAs are formed by the vertical connection of the first seven cervical intersegmental arteries. The horizontal segments of the cervical intersegmental arteries are connected to the dorsal aorta. During the development of the horizontally positioned cervical intersegmental arteries, all but the proximal part of the SCA and the 7th cervical intersegmental artery, which forms the origin of the VA, disappear.[8] However, it is suggested that the connection of the right VA with the right SCA is lost due to obliteration of the distal right dorsal aorta segment between the 6th and 7th cervical intersegmental arteries during the formation of VAL anomaly. The right SCA normally develops from the 7th cervical intersegmental artery, but the right VA is thought to originate from the persistent proximal right dorsal aorta distal to the left SCA [Figure 3]. There are publications claiming that the Kommerell's diverticulum seen in the right VA origin, which we detected in our case, is the remnant of this right dorsal aortic origin.[8,9]

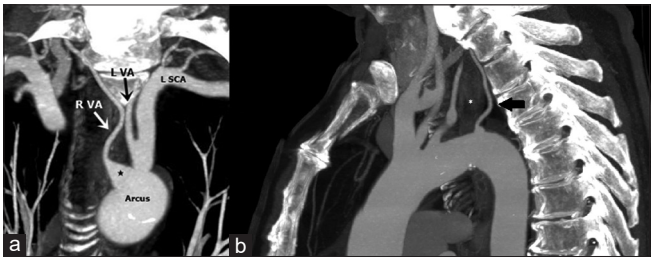

Figure 1: (a) The coronal CT maximum intensity projection image showed Kommerell's diverticulum (*) in the origin of the RVA and the extreme proximal origin of the LVA from the LSCA (arrow). (b) vThe sagittal maximum intensity projection CT image showed the retroesophageal course of the aberrant RVA. RVA: Right vertebral artery, LVA: Left vertebral artery, LSCA: Left subclavian artery, CT: Computed tomography

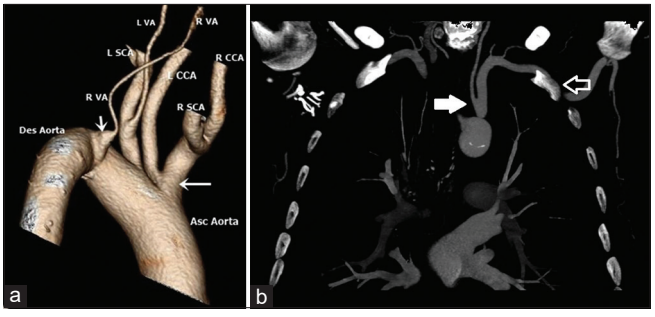

Figure 2: (a) a-The dorsal oblique 3D CT image showed the RVA with Kommerell's diverticulum in its origin, arising from the dorsal aspect of the aortic arch and distal to the LSCA (small arrow). Also, bovine arch was seen (large arrow). (b) The coronal maximum intensity projection CT image showed that the origin of the LVA (white arrow) was below the first rib (open arrow). Des Aorta: Descending aorta, LVA: Left vertebral artery, LCCA: Left common carotid artery, RCCA: Right common carotid artery, RSCA: Right subclavian artery, Asc Aorta: Ascending Aorta, RVA: Right vertebral artery, LSCA: Left subclavian artery, CT: Computed tomography

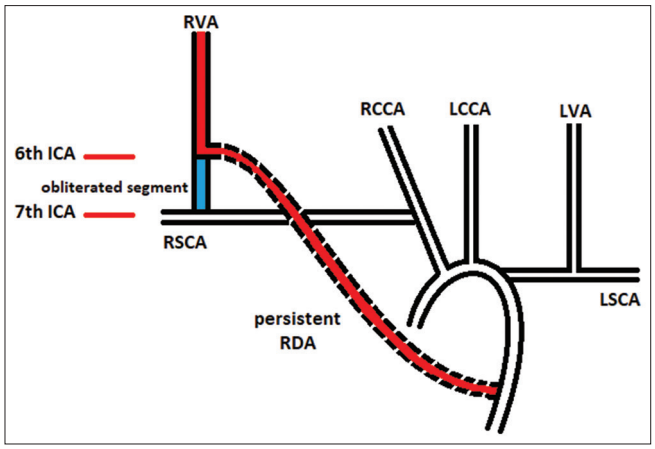

Figure 3: A simple scheme of the embryological development of vertebral arteria lusoria. Due to the obliteration of the vertical segment between the 6th and 7th intersegmental arteries, the connection between the RVA and the RSCA get lost. Instead, the RDA does not regress and provides the continuity of the RVA with aortic arch. ICA: Cervical Intersegmental Artery, RCCA: Right Common Carotid Artery, LCCA: Left Common Carotid Artery, LVA: Left Vertebral Artery, LSCA: Left Subclavian Artery, RDA: Right dorsal aorta, RSCA: Right subclavian artery, RVA: Right vertebral artery

Few cases in the literature have been reported as symptomatic. VAL may compress the esophagus or trachea as in dysphagia lusoria due to aberrant SCA. Apart from dysphagia, since the VA is thinner than the SCA, headache and dizziness may be rarely seen due to the influence of the VA that feeds the cerebral system.[9] We think these symptoms are less common thanks to the compensation mechanisms (posterior communicating arteries) between the posterior and anterior vascular circulations of the brain and the support of the opposite VA in the posterior circulation. However, it has been reported that variations in the great vessels of the aortic arch may adversely affect hemodynamics and predispose to vascular pathologies such as dissection, aneurysm, and arteriovenous fistula.[7,9] Our case did not have any symptoms related to these anomalies or a vascular pathology detected by imaging methods.

VAL may be detected incidentally by catheter angiography examination or encountered in multislice CT examinations, which are more frequently used in daily practice. In contrast to the right aberrant SCA, the VAL is thinner and may be overlooked by the radiologist. Failure to draw attention to this anomaly by the radiologist may lead to life-threatening neurological complications such as large hemomediastinum or stroke due to unstoppable hemorrhages due to arterial damage during esophageal surgeries, minimally invasive endoscopic procedures, or interventional endovascular procedures that are increasing nowadays.[8]

Conclusion

As a result, we may encounter vascular anomalies of the aortic arch incidentally during cross-sectional radiological imaging. Furthermore, other rare variations may accompany rare anomalies such as VAL. It is important to keep these in mind and draw attention to them, as it will reduce the wrong evaluation and complications that may occur during surgical, endoscopic, or interventional endovascular procedures.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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