



# Vertebral artery triplication

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## Abstract

**Purpose** To report an extremely rare vertebral artery (VA) origin configuration.

**Methods** Angiographic and MRI features of a VA triplication in a child are presented.

**Results** The VA trifurcation was the result of the simultaneous persistence of the 4th, 5th and 6th primitive cervical intersegmental arteries, respectively, reaching the transverse canal at C4, C5 and C6. A contralateral VA duplication was noted as well.

**Conclusion** A previously undocumented instance of VA triplication is presented. Knowledge of such variations is important for the planning of diagnostic and therapeutic cervicocranial angiography and the understanding of unusual images on noninvasive imaging studies. This type of anomaly also represents a risk factor for arterial dissection.

**Keywords** Anatomical variation · Arteries · Vertebral artery · Angiography

## Introduction

The adult vertebral artery (VA) combines vascular elements of separate embryonic origins: the cranial portion derives from the primitive pro-atlantal artery of Padget–Arey [8], the caudal portion from a succession of anastomoses linking the first six primitive cervical intersegmental arteries (ISA) [4]. As the cranial and caudal components connect to form the VA, the aortic stems of the primitive cervical ISAs C1–C5 involute while C6 becomes part of the subclavian artery [8]. The persistence of the aortic stem of a primitive cervical ISA at a level other than C6 constitutes a variation known as a persistent cervical ISA.

The persistence of a single primitive cervical ISA results in an anatomical configuration known as an aberrant VA origin, for example, from the aortic arch or from a carotid artery. When two primitive cervical ISAs persist simultaneously, the VA appears to have a dual origin: the variant is thus called a VA duplication.

The persistence of more than two primitive cervical ISAs is exceptional. In his classic work on the arteries of

the human body (1844) [10], Quain described a trifurcated VA but his variant was not made of three persistent cervical ISAs (Fig. 1). No other observation—anatomical or radiological—of VA trifurcation has to our knowledge been reported. We present here a case of VA triplication in a child.

## Case report

The variant was incidentally observed in a 3-year-old boy undergoing angiography during the investigation and treatment of a pharyngeal hemangioma. The right VA had three separate origins from the right subclavian artery, respectively, corresponding to persistent 4th, 5th and 6th ISAs (Fig. 2A–D). The left VA was duplicated, the proximal segment being made of 4th and 6th persistent cervical ISAs. There was no other documented variation. The hemangioma was embolized in preparation for surgical resection.

## Discussion

### Vertebral artery development

The cervical and cranial portions of the VA follow dissimilar and asynchronous developmental patterns [4]. The cranial segment essentially derives from the pro-atlantal artery,

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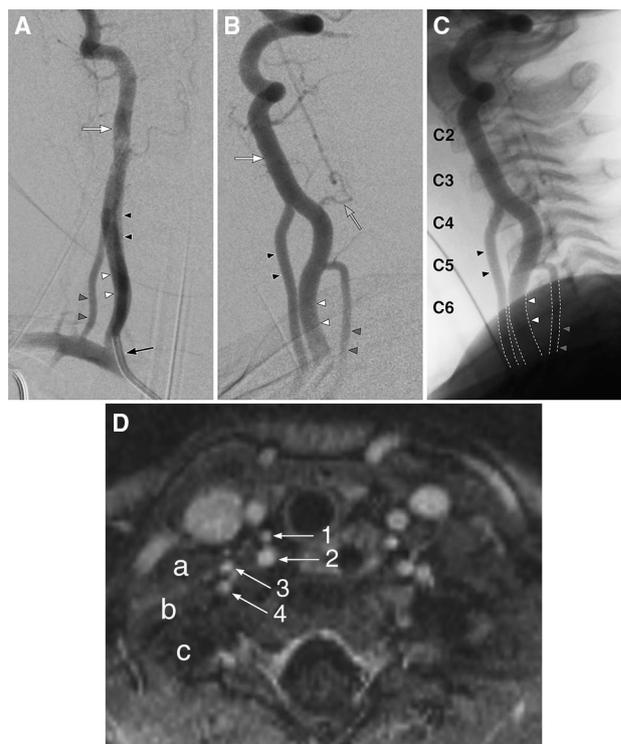
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**Fig. 1** Illustration reproduced from Quain's *The anatomy of the arteries of the human body, with its applications to pathology and operative surgery* (1844) (author's collection). In Quain's words, "in this figure is represented a very unusual arrangement of the right vertebral artery which results from the union of three branches". The original legend indicates that VA (6) has three origins (6a, 6b, 6c), the last one being a branch of the inferior thyroid artery (7), i.e., the ascending cervical artery. The other two origins correspond to persistent cervical ISAs (4th and 6th), identified by their level of passage into the transverse canal. Anastomoses between the VA and the ascending cervical artery are not subjected to this rule and can, as shown here, be established at any vertebral levels

remnants of which participate in the formation of the vertebrobasilar junction [1]. The spinal branch, in particular, provides a prominent radicular artery that courses along the first cervical nerve and becomes the terminal segment of the adult VA [3].

The cervical portion of the VA, on the other hand, consists of a succession of longitudinal anastomoses established between the first 6 cervical ISAs. This anastomotic chain normally remains connected caudally to the subclavian artery and cranially to the pro-atlantal artery [8].



**Fig. 2** Three-year-old boy with pharyngeal hemangioma. **A** DSA, right vertebral artery angiography, posteroanterior projection. This injection was performed by selective catheterization (black arrow) of the most proximal of the three VA origins, i.e., the persistent 4th cervical ISA; the other two persistent ISAs are opacified retrogradely. In this projection, the 4th (black arrowheads) and 5th (white arrowheads) cervical ISAs are partially projecting over each other but can still be identified as separate vessels. The third limb, more distal, is well appreciated (grey arrowheads). The three proximal segments coalesce into a single right VA above C4 (white arrow). **B** DSA, right vertebral artery angiography (same injection), lateral projection. All three persistent cervical ISAs are clearly identified in this projection: 4th (black arrowheads), 5th (white arrowheads) and 6th (grey arrowheads), continuing cranially as a single right VA (white arrow). Note the presence of a prominent right anterior radiculomedullary artery at C4 (grey arrow). **C** DSA, right vertebral artery angiography, lateral projection (same as B, non-subtracted image), offering topographic correlations with osseous landmarks. The point of entrance into the transverse canal and the most caudal transverse foramen crossed allow to identify with certainty which primitive cervical ISA persists at the adult stage, in this case the 4th (black arrowheads), 5th (white arrowheads) and 6th (grey arrowheads) cervical ISAs. **D** MRA, axial plane at C7, confirming the topography of the three persistent cervical ISAs (1=4th ISA, 2=5th ISA, 3=6th ISA) deep to the scalene muscle (**A**, **B** and **C**, respectively, indicate anterior, median and posterior scalene muscles). The right vertebral vein is also visible (4). If one of the limbs of the VA triplication was made of the ascending cervical artery (as in Fig. 1), it would lie superficial to the scalene muscle

## VA duplications and persistent cervical ISAs

The site of origin of the VA depends on the cervical ISA that persists at the adult stage to become its proximal (V1) segment. While the latter generally derives from the 6th cervical ISA, any cervical (or upper thoracic) ISA can become the proximal adult VA. This persistent vessel is identified by its level of entrance into the transverse canal rather than by its origin. For example, a VA formed by a persistent 5th cervical ISA always enters the C5 transverse foramen but may originate from the subclavian artery, from the aortic arch between the left common carotid and subclavian arteries, or even from a common carotid artery.

Persistent cervical ISAs (other than the one becoming the V1 segment) are rare. To our knowledge, there is no published case of true persistent 1st ISA. Persistent 2nd cervical ISAs—running along the 3rd cervical nerve root between C2 and C3—are exceptional, with only two observations reported so far [2, 9]. Several cases of “persistent 1st ISA pro-atlantal artery” have been published; these reports generally claim to follow Padget’s classification even though she clearly stated that using the name of 1st ISA for the pro-atlantal artery—one of the carotid–vertebral anastomoses—was “indefensible” [8]. Persistent 3rd, 4th and particularly 5th cervical ISAs are more common, notably in a context of proximal VA duplication. The second limb of the duplication is usually (but not always) made of the 6th cervical ISA.

While the developmental history of the VA can, therefore, easily account for a triple origin of that vessel—or VA triplication—there is to our knowledge no prior documentation of this variation. Quain reported an instance of triplicated VA in 1844 but his observation was slightly different, as only two limbs corresponded to persistent primitive cervical ISAs (i.e., 4th and 6th), the most distal one being an anastomosis with the ascending cervical artery [10]. In our case, the left VA triplication is made of the 4th, 5th and 6th persistent cervical ISAs, respectively, passing through the C4, C5 and C6 transverse foramina.

A persistent cervical ISA—either as an isolated variant or as part of a vertebral duplication or triplication—might be a risk factor for arterial dissection [5, 6]. The abrupt curve of the vessel immediately prior to its entrance into

the transverse canal represents a transition between mobile and fixed segments that can be stretched and/or compressed, notably during neck rotation. Conversely, a VA duplication may be confused with an arterial dissection [7].

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## Compliance with ethical standards

**Conflict of interest** P. Gailloud is a consultant for Cerenovus (honoraria) and a recipient of research Grants from Siemens Medical (paid to institution).

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