

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

Anomalous external carotid-internal carotid artery anastomosis diagnosed by computed tomography angiography: a case report ☆,☆☆,★

Byung Hoon Lee, MD*

Department of Radiology, Inje University Ilsan Paik Hospital, 170 Juhwaro, Ilsanseogu, Goyang, Gyeonggido, South Korea

ARTICLE INFO

Article history:

Received 4 July 2020

Revised 24 July 2020

Accepted 25 July 2020

Keywords:

Anomalous anastomosis

Embryology

External carotid artery

Internal carotid artery

Nonbifurcating carotid artery

Occipital artery

ABSTRACT

An anomalous anastomosis between the external and internal carotid arteries forming an arterial ring at the level of the cervical internal carotid artery is an extremely rare variant, and its embryological origin remains unclear. This report describes a rare arterial variation at the level of carotid bifurcation incidentally discovered in a 67-year-old man. Computed tomography angiography revealed an anomalous anastomosis between the external and internal carotid arteries at the level of the C2–3 intervertebral space with a small proximal cervical segment of the internal carotid artery and the occipital artery originating at this anomalous anastomosis. This report may be of embryological significance and contribute to understanding the mechanisms of carotid artery formation.

© 2020 The Authors. Published by Elsevier Inc. on behalf of University of Washington.

This is an open access article under the CC BY-NC-ND license.

(<http://creativecommons.org/licenses/by-nc-nd/4.0/>)

Introduction

The common carotid artery (CCA) typically divides into the internal carotid artery (ICA) and external carotid artery (ECA) at the C3–4 vertebral level [1]. A nonbranching CCA is a rare anatomical variation, in which the major branch of the proximal ECA directly arises from the common trunk without forming a carotid bifurcation [1,2]. However, its etiology

remains unclear [1]. A congenital anastomosis between the ECA and ICA forming an arterial ring at the cervical ICA level is exceedingly rare and seems to be a nonbranching CCA variant [3]. I herein report a case of an anomalous artery directly connecting the ECA and ICA at the level of the C2–3 intervertebral space with hypoplastic proximal cervical ICA diagnosed by computed tomography angiography (CTA). In addition, the embryological mechanism underlying the development of this rare vascular variation is discussed.

☆ Funding: There was no funding support for the research.

☆☆ Declaration of Competing Interest: The author declare that they have no conflict of interest.

★ This study has got an ethical review exemption from the Institutional Review Board of author's institution, which waived the requirement for written informed consent from the participant. All radiologic images are entirely anonymized.

* Corresponding author.

<https://doi.org/10.1016/j.radcr.2020.07.072>

1930-0433/© 2020 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license. (<http://creativecommons.org/licenses/by-nc-nd/4.0/>)

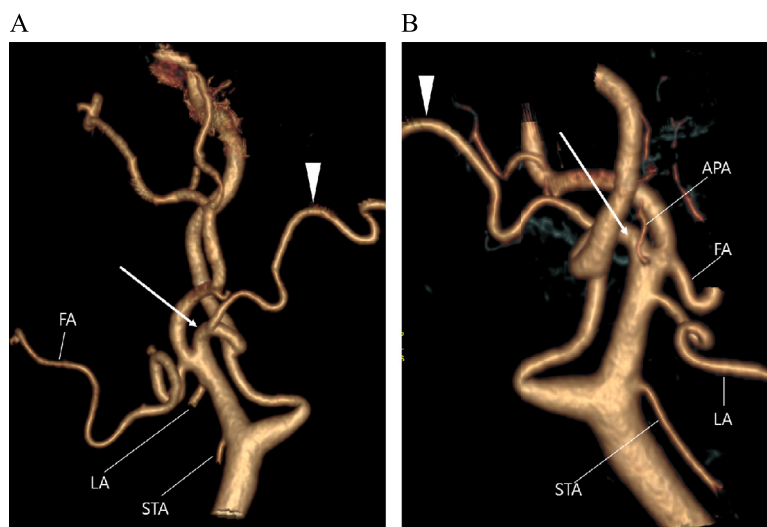


Fig. 1 – CT angiography images. Three-dimensional volume-rendered CT angiography of the neck revealed an anomalous external carotid artery–internal carotid artery anastomosis (white arrow) at the level of the C2–3 intervertebral space. The occipital artery (white arrowhead) originated from the anomalous anastomosis. APA, ascending pharyngeal artery; FA, facial artery; LA, lingual artery; STA, superior thyroid artery.

Case report

A 67-year-old male patient was admitted to our emergency department with a 4-hour history of dysarthria and right-sided hemiparesis. CTA was performed to determine the presence or absence of large artery occlusion at admission. Carotid bifurcation and the carotid bulb were observed bilaterally at the C4 vertebral body level. The proximal cervical segment of the left ICA was small in caliber and the main trunk of the ECA was relatively large. An anomalous anastomosis between the ICA and ECA was observed at the level of the C2–3 intervertebral space, forming an arterial ring at the proximal cervical ICA; the left occipital artery originated from this anomalous connecting artery (Figs. 1 and 2). The ICA distal to the anomalous anastomosis was normal in caliber and ascended into the carotid canal to become the petrosal segment of the ICA. The intracranial course of the bilateral ICA and the vertebrasilar system were typical in appearance. The CTA source images revealed a normal bilateral carotid canal. There was neither evidence of a persistent stapedia artery nor enlargement of the inferior tympanic canaliculus. However, 1 hour later, diffusion-weighted imaging and the corresponding apparent diffusion coefficient map revealed restricted diffusion in the left thalamus (Fig. 3). Magnetic resonance angiography also demonstrated an anomalous ECA–ICA connection with a hypoplastic proximal cervical ICA (Fig. 4).

Discussion

Embryologically, the ICA develops from the third aortic arch and the dorsal aortic roots cranial to the third aortic arch during early gestation [4,5]. In contrast, the ECA develops from 2

connected arteries [4,6]. The ventral aortic roots cranial to the third aortic arch become the ventral pharyngeal artery and the ventral second aortic arch finally regresses [5]. The dorsal second aortic arch becomes the hyoid artery. The stapedia artery, the main branch of the hyoid artery, connects the ventral pharyngeal artery and separates from the hyoid artery at the 24 mm embryonic stage [5,7]. The typical ECA then derives from the combination of the ventral pharyngeal artery and stapedia artery [5].

Nonbranching CCA is a rare congenital anatomical variation of the cervical carotid artery [1,2]. Both nonbranching CCA and anomalous ECA–ICA anastomosis forming an arterial ring seem to be similar developmental variations [1,3]. Two hypotheses have been proposed to explain the causes of a nonbranching CCA [2,4]. I assumed that a similar hypothesis could be applied in the present case.

The first theory explaining the formation of a nonbranching CCA is agenesis of the main trunk of the ECA [3,4]. In this situation, the persistence of the hyoid–stapedia artery system would be the main supply of the ECA territory [3]. In the present case, however, this hypothesis was not applicable because the main trunk of the ECA was large; the main branches, such as the facial and lingual arteries, arose from the typical location, whereas the proximal cervical ICA was small. In addition, there was no persistent stapedia artery on the CTA source images.

The second hypothesis is agenesis of the proximal segment of the ICA [6]. In some cases, an arterial stump in the proximal cervical ICA has been observed, which would support this second hypothesis [1,6–8]. Kim et al. reported 2 cases of anomalous ECA–ICA anastomosis with proximal ICA remnants or hypogenesis, and suggested that a nonbranching CCA results from the agenesis or hypogenesis of the proximal ICA with normal development of the main ECA trunk [1]. Like the report by Kim et al., in the present case, the proximal cervi-

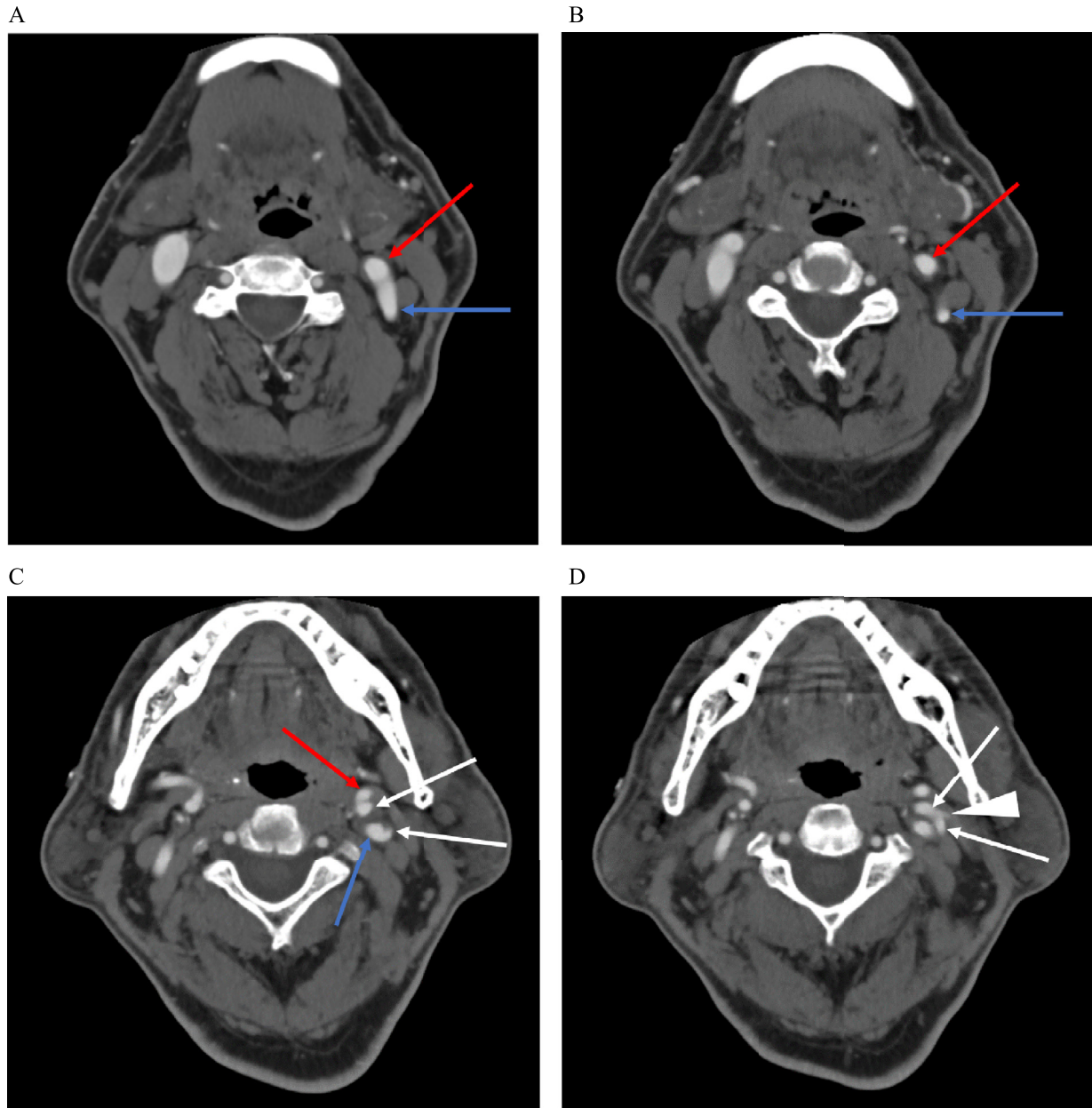


Fig. 2 – Axial CT images. (A) Axial CT demonstrated left carotid bifurcation at the level of the C4 vertebra. The main trunk of the ECA (red arrow) and proximal cervical segment of the ICA (blue arrow) can be seen just distal to the bifurcation. **(B)** The main trunk of the ECA (red arrow) and the small size of the proximal cervical segment of the ICA (blue arrow) at the level of the C3–4 intervertebral space. **(C)** The anomalous anastomosis (white arrows) connecting to the ECA (red arrow) and ICA (blue arrow) at the level of the C2–3 intervertebral space. **(D)** The left occipital artery (white arrowhead) originating from the anomalous anastomosis (white arrows) at the level of the C2 vertebra.

cal ICA had a small diameter and the main trunk of the ECA was relatively large, thus supporting the hypothesis of agenesis of the proximal cervical ICA. The anomaly in this case is considered to result from hypogenesis of the proximal cervical segment of the ICA. If the small proximal ICA was either occluded or did not form during the gestational period, this anomaly may have the same characteristics as a nonbranching CCA. Therefore, this anatomical variation of an anomalous ECA-ICA anastomosis forming an arterial ring seems to be a

nonbranching CCA variant. In addition, this anatomical variation at the proximal cervical segment can be thought of as an intermediate step between normal carotid bifurcation and nonbranching CCA development.

When abnormal regression of the third aortic arch occurs, the distal ICA is constructed via a variable anatomical network as a collateral pathway [3,9]. Abnormal regression of the third aortic arch might result in the persistence of the hyoid artery and could produce high bifurcation of the carotid artery in a

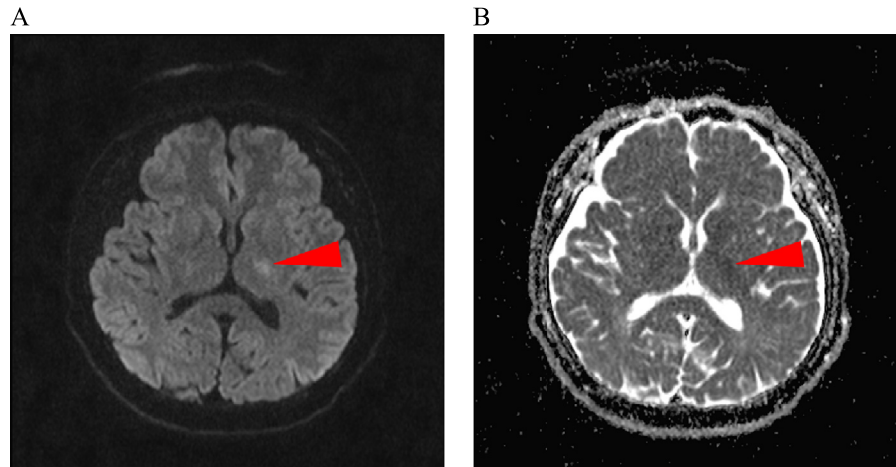


Fig. 3 – Axial diffusion-weighted image (A) and the corresponding apparent diffusion coefficient map (B) showed restricted water diffusion in the left thalamus (red arrowheads).

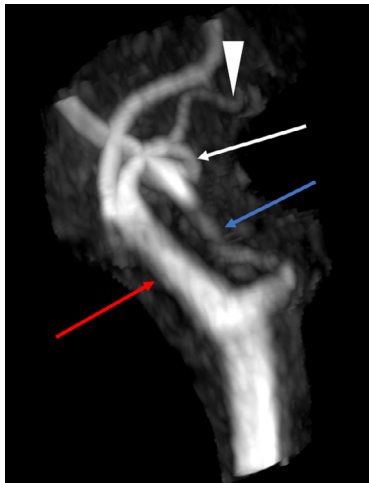


Fig. 4 – Time-of-flight magnetic resonance angiography of the neck revealed the anomalous anastomosis (white arrow) between the ECA (red arrow) and ICA (blue arrow). The occipital artery is shown (white arrowhead) arising from the anomalous connection.

nonbranching CCA [10]. The tortuosity of the nonbranching CCA at the level of the C1 vertebra suggests that the primitive ECA and ICA are connected [2,4]. However, the present case exhibited carotid bifurcation at the C3–4 vertebral level and anomalous anastomosis between the ICA and ECA at the level of the C2–3 intervertebral space. Therefore, it can be assumed that in this case there existed a pathway other than the collateral through the second aortic arch.

The occipital artery typically arises from the posterior aspect of the proximal ECA and courses posteriorly [11]. Lasjaunias et al. suggested that the occipital artery forms from the proatlantal artery; can arise from the primitive ICA, ECA, and vertebral artery; and may be determined by the sites of regression [12]. In early gestation, the proatlantal artery anas-

tomoses with primitive arteries, and the anastomotic vessel between the primitive ICA and proatlantal artery generally regresses, resulting in the formation of a typical occipital artery of ECA origin [11,12]. Persistence of anastomotic vessels between the proatlantal artery and the primitive ICA, and regression of the anatomical connections between the proatlantal artery and the primitive ECA, can result in the formation of an occipital artery of ICA origin [11,12]. In a similar manner, when there is a persistent anatomical connection between the proatlantal artery and the primitive ICA, and the proximal segment of the proatlantal artery from the primitive ECA does not regress, the occipital artery can arise from the anomalous anastomosis between the proximal ICA and ECA (Fig. 5). In the present case, the left occipital artery, which is typically derived from the proatlantal artery as a branch of the ventral pharyngeal artery, originated from the anomalous connecting artery between the ECA and ICA. Sasaki et al. suggested that a nonbranching CCA is formed by agenesis of the proximal part of the cervical ICA using the proatlantal artery or hypoglossal artery as a collateral pathway [7]. Kim et al. also reported an anatomical variation with an anomalous anastomosis at the C2–3 spinal level, and the occipital artery originating from this anomalous vessel, as detected by catheter angiography [1]. However, there was no abnormality in any of the derivative arteries of the hyoid-stapedial artery system from the second aortic arch, such as the middle meningeal or caroticotympanic arteries. Thus, regarding embryological development, it is plausible that the present anomalous artery may be derived from the proatlantal artery, which is normally derived from the occipital artery. In this case, the proximal occipital artery likely plays an important role in forming an arterial ring at the proximal cervical ICA.

The ICA reconstructed via the second aortic arch is known as an aberrant ICA. It is generally thought that an aberrant ICA is formed from the anastomosis between the inferior tympanic artery and caroticotympanic artery, a hyoid artery remnant that is derived from the second aortic arch [7]. However, the anomalous anastomosis in the present case exhibited no aberrant course of the ICA or enlarged inferior

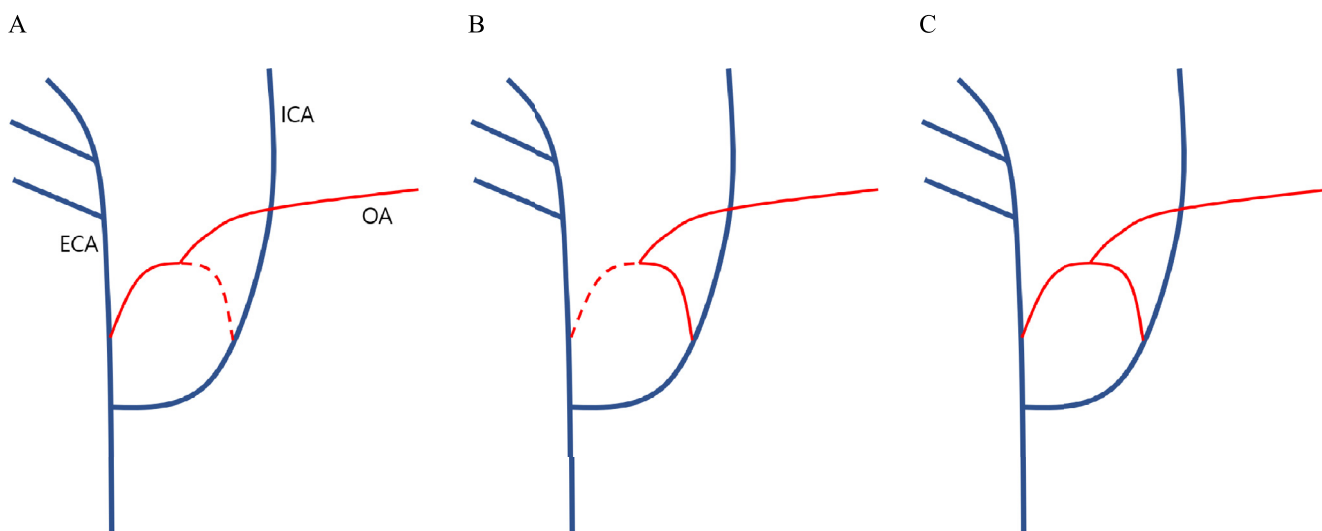


Fig. 5 – Schematic illustration of the speculated embryogenesis of the present case in a lateral projection (modified from Uchino et al. [11]). (A) Normal development of a typical occipital artery (OA). Dotted lines are regressed arteries. (B) OA arising from the internal carotid artery (ICA). (C) Present case. The anomalous anastomosis between the external carotid artery (ECA) and ICA, and the OA originating from the anomalous anastomosis.

tympenic canaliculus. The anatomical variation in this study differs from acquired carotid dissection, fenestration, or duplication because the 2 different arteries form an arterial ring with the anomalous ECA-ICA anastomosis.

Conclusion

This report describes an extremely rare case of anomalous ECA-ICA anastomosis forming an arterial ring at the cervical segment, with the occipital artery originating from this anomalous connection, as visualized by CTA. This case provides evidence for one of the possible etiologies of the formation of a nonbranching CCA, and may indicate some embryological mechanisms underlying the formation of the carotid arteries. Clinically, this case also indicates that careful review of angiography findings is important to identify such rare arterial variations at the carotid bifurcation level to avoid complications during either head and neck surgeries or endovascular procedures.

Author contribution

Byung Hoon Lee; design of the study, patient enrollment, data acquisition, data analysis/interpretation, and manuscript preparation.

REFERENCES

- [1] Kim CH, Cho YD, Kang HS, Kim JE, Jung SC, Ahn JH, et al. Anomalous external carotid artery-internal carotid artery anastomosis in two patients with proximal internal carotid arterial remnants. *Korean J Radiol* 2015;16(4):914–18 PubMed:26175593]. doi:10.3348/kjr.2015.16.4.914.
- [2] Yoshida S, Ota T. Carotid endarterectomy for atherosclerotic stenosis associated with non-bifurcating cervical carotid artery: a case report. *NMC Case Rep J* 2016;3(3):59–62 PubMed:28663999]. doi:10.2176/nmccrj.cr.2015-0232.
- [3] Nakai K, Kaji T, Uchino A, Kawachi T, Tamura C, Otani N, et al. Congenital external carotid-internal carotid artery anastomosis associated with contralateral non-bifurcating cervical carotid artery. *Neuroradiology* 2012;54(5):521–3 PubMed:21922207]. doi:10.1007/s00234-011-0956-x.
- [4] Ooigawa H, Nawashiro H, Fukui S, Tsuzuki N, Katoh H, Kawaguchi T, et al. Non-bifurcating cervical carotid artery. *J Clin Neurosci* 2006;13(9):944–7 PubMed:17052907]. doi:10.1016/j.jocn.2005.10.015.
- [5] Hu CC, Lai YJ, Lai WJ. Nonbifurcating carotid artery: a case report with a review of embryogenesis. *Am J Case Rep* 2018;19:891–5 PubMed:30057401]. doi:10.12659/AJCR.910019.
- [6] Muro S, Nakanishi Y, Minami T, Matsui O, Furukawa M, Yoshizaki T. Case report. Intra-arterial chemotherapy for laryngeal cancer via a non-bifurcating carotid artery. *Br J Radiol* 2009;82(982):e197–9 PubMed:19759204]. doi:10.1259/bjr/78301422.
- [7] Sasaki T, Nagashima H, Oya F, Satoh D, Kobayashi S. Carotid artery stenting for atherosclerotic stenosis associated with non-bifurcating cervical carotid artery. *Neurol Med Chir (Tokyo)* 2013;53(4):228–32 PubMed:23615413]. doi:10.2176/nmc.53.228.
- [8] Yuan C, Duan H, Yi Z, Zhang Y, Zhang J. Stenting for middle cerebral artery stenosis associated with a nonbifurcating cervical carotid artery: case report and naming consideration. *World Neurosurg* 2019;125:170–4 PubMed:30763749]. doi:10.1016/j.wneu.2019.01.208.
- [9] Uchino A, Saito N, Watadani T. Congenital external carotid-internal carotid artery anastomosis diagnosed by MR angiography. *J Neuroimaging* 2013;23(1):96–7 PubMed:21699608]. doi:10.1111/j.1552-6569.2011.00613.x.
- [10] Uchino A, Saito N, Watadani T, Mizukoshi W, Nakajima R. Nonbifurcating cervical carotid artery diagnosed by MR

[1] Kim CH, Cho YD, Kang HS, Kim JE, Jung SC, Ahn JH, et al. Anomalous external carotid artery-internal carotid artery

- angiography. *AJNR Am J Neuroradiol* 2011;32(6):1119–22 PubMed:21622581]. doi:[10.3174/ajnr.A2462](https://doi.org/10.3174/ajnr.A2462).
- [11] Uchino A, Saito N, Mizukoshi W, Okada Y. Anomalous origin of the occipital artery diagnosed by magnetic resonance angiography. *Neuroradiology* 2011;53(11):853–7 PubMed:21174081]. doi:[10.1007/s00234-010-0825-z](https://doi.org/10.1007/s00234-010-0825-z).
- [12] Lasjaunias P, Theron J, Moret J. The occipital artery. Anatomy–normal arteriographic aspects–embryological significance. *Neuroradiology* 1978;15(1):31–7 PubMed:643171]. doi:[10.1007/BF00327443](https://doi.org/10.1007/BF00327443).