

A Rare and Subtle Etiology of Chronic Oropharyngeal Pain: Isolated Internal Carotid Artery Kinking without Stenosis

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Carotid artery kinking is a frequent finding in duplex ultrasonography. However, isolated morphological changes without significant carotid stenosis are rarely symptomatic. Neck pain is a rare symptom in patients with carotid artery kinks. The vascular etiology in patients with persistent neck pain is usually overlooked. A 58-year-old female patient with chronic neck pain presented to our clinic. Following multidisciplinary team review, the symptoms were found due to the kinking of the internal carotid artery. In this report, we present the clinical presentation of the patient with the kinking of the internal carotid artery without stenosis, surgical management of the pathology, and a brief literature review.

Morphological changes in internal carotid artery such as kinking are frequent among the general population; however, it is asymptomatic in most of the cases. Depending on asymptomatic population, carotid artery kinking is usually diagnosed incidentally. It has an increased incidence rate in older age group, female gender, patients with hyperlipidemia, hypertension, diabetes mellitus, and ischemic heart disease.¹

Despite being a silent disorder in most of the cases, internal carotid artery kinking without significant stenosis may result in hemispheric and nonhemispheric symptoms. There are controversial data

on whether morphological changes in internal carotid artery may cause carotid artery stenosis and ischemic stroke.² In cases of cerebral insufficiency, carotid artery screening is almost always required, which reveals the abnormal course of the carotid artery. However, in patients with nonhemispheric symptoms such as chronic neck pain, the vascular etiology is usually overlooked.

In this case report, we present a 58-year-old female patient who had chronic neck pain which could not be relieved despite various surgical interventions. Multidisciplinary team review provided no other cause for the symptoms other than internal

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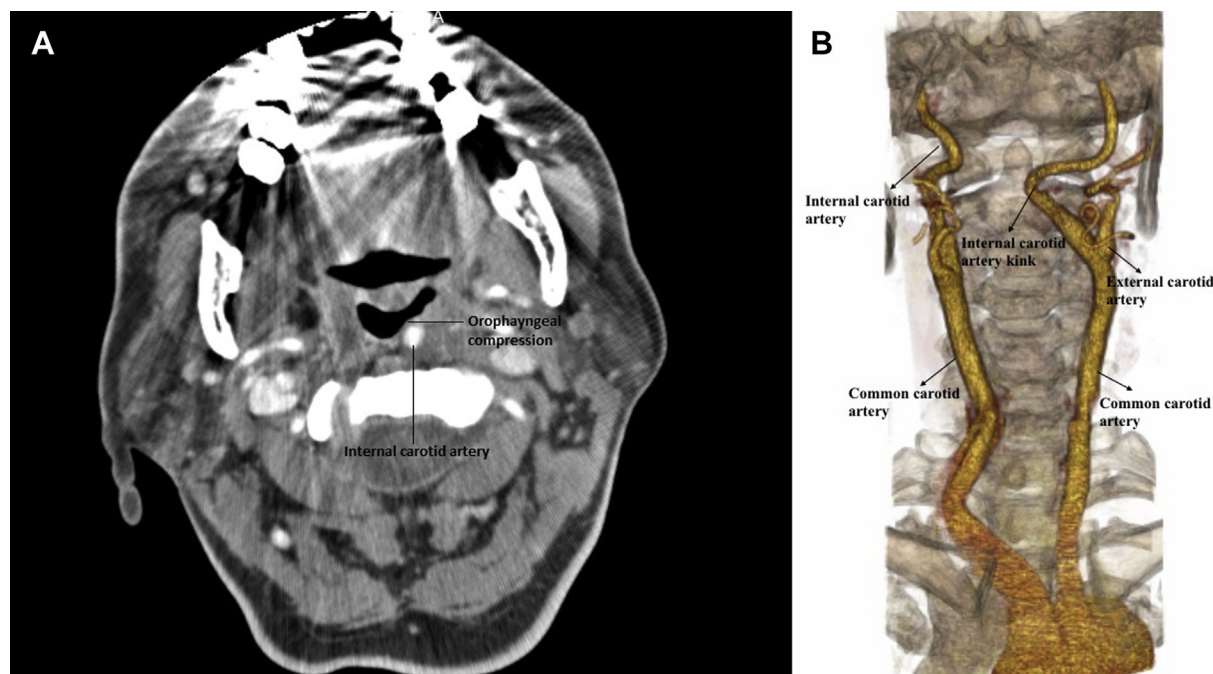


Fig. 1. (A) Preoperative computerized tomography angiography showing *left* internal carotid artery kink, medial deviation, and oropharyngeal compression. (B)

Preoperative computerized tomography angiography showing *left* internal carotid artery kink and medial deviation.

carotid artery kinking. Hence, the patient was considered for surgical correction.

CASE REPORT

A 58-year-old female patient was referred to our clinic with chronic pain at the oropharynx. Physical examination revealed a pulsating mass and ulceration at the left posterolateral side of the oral cavity above the left tonsil. Manual compression over the eroded region increased the pain of the patient. Auscultation of the neck indicated an atypical bruit over the left carotid artery. The patient has previously been investigated for the pain and underwent bilateral tonsillectomy and thyroidectomy; however, her symptoms persisted.

Carotid artery duplex ultrasonography showed a kinked left internal carotid artery without stenosis. Computed tomography angiography confirmed the tortuosity of the left internal carotid artery with increased deviation toward the oropharynx (Fig. 1). The patient was consulted with neurology and ear-nose-throat departments and no other cause for the pain symptoms was identified. She was scheduled for surgical treatment after her consent.

Operation was performed with general anesthesia. Neurologic monitoring was provided with continuous near infrared spectromicroscopy (NIRS) evaluation throughout the procedure. Following left cervical

incision, common carotid, internal and external carotid arteries were dissected and looped. There was medial kink at the superior segment of the internal carotid artery (Fig. 2A). The internal carotid artery was long segment dissected free from adhesions from the adjacent structures. The carotid arteries were clamped after systemic 100 U/kg heparin administration. The internal carotid artery was divided from the carotid bifurcation. It was retracted approximately 2.5–3 cm inferiorly (Fig. 2B) and reanastomosed to the common carotid artery (Fig. 3B). The internal carotid artery stump was primarily sutured and clamps were removed. A significant decline in the NIRS values was not observed throughout the surgery and the patient was extubated without any neurologic deficit immediately. Postoperative course was uneventful, her symptoms disappeared, and pulsating image at the left oropharynx was not observed. She was discharged from the hospital on the postoperative third day. She has been followed symptom free and postoperative sixth-month and first-year control computerized tomography angiography indicated normally configured carotid arteries away from the oropharynx (Figs. 3 and 4, cine).

DISCUSSION

The etiology of the morphological changes in internal carotid artery remains controversial. It is

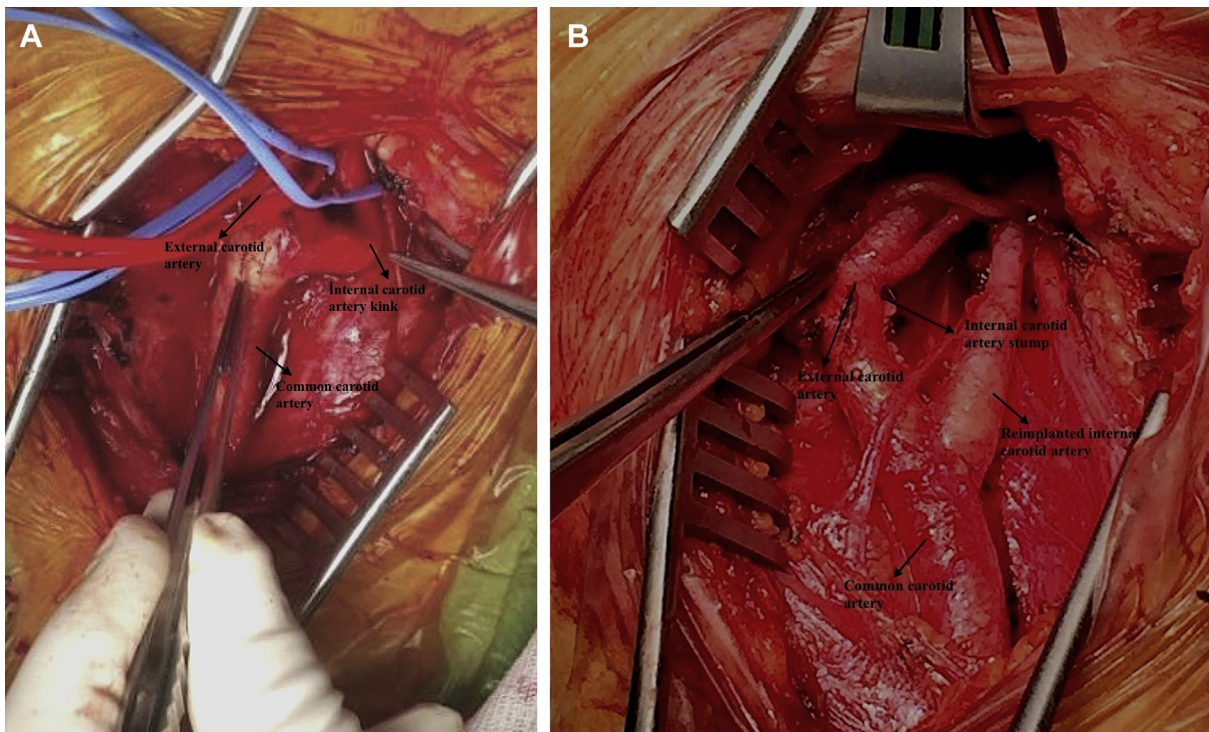


Fig. 2. Surgical exploration and treatment. **(A)** Surgical view of the kink. (The internal carotid artery is retracted for a better quality image). **(B)** Resection, retraction, and

proximal reimplantation of the internal carotid artery to the common carotid artery.

mentioned that fibromuscular dysplasia has a possible etiological role. Another hypothesis is that kinking of internal carotid artery can be congenital, resulting from embryological causes.³ Even though



Fig. 3. Postoperative sixth-month control computerized tomography angiography.

the pathophysiological mechanism of the morphological changes in the internal carotid artery remains unclear, it is known that carotid kinking has an increased incidence rate in older age group, female gender, and patients with hyperlipidemia, hypertension, diabetes mellitus, and ischemic heart disease.¹ In our case, the patient was female, had a history of diabetes mellitus and hypertension, hence she was in consistency with the presumed risk factors.

There is surgical indication to repair the significantly stenotic kinking of the internal carotid artery in patients with neurologic symptoms. However, the data are controversial on treatment of the isolated morphological changes without carotid stenosis. This controversy is based on whether isolated morphological changes in internal carotid artery cause cerebral insufficiency or not.² In the “2017 Clinical Practice Guidelines of the European Society for Vascular Surgery,” it is stated that surgical intervention for asymptomatic isolated coils/kinks of the internal carotid artery is not recommended. However, symptomatic patients with isolated coils/kinks that provided no other cause for symptoms may be considered for surgical correction following multidisciplinary team review, as mentioned in the guideline with IIb class of recommendation.⁴ We

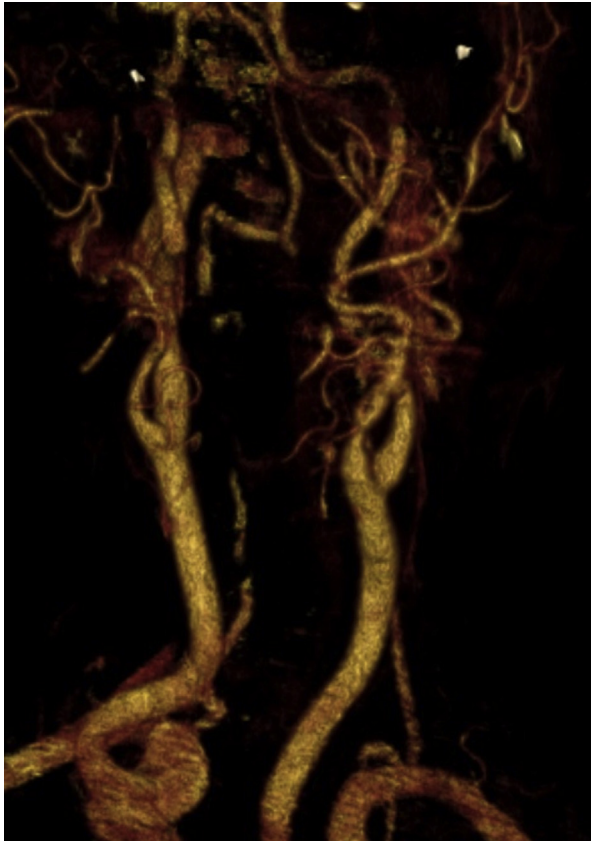


Fig. 4. Postoperative first-year control computerized tomography angiography.

consulted our particular patient with neurology and ear-nose-throat departments due to persistent neck pain and no other cause for the pain symptoms could be identified other than isolated carotid kinking. Therefore, the patient was scheduled for surgical treatment.

The treatment of the morphological changes in internal carotid artery may be performed with various techniques. Resection and end-to-end anastomosis, shortening and reimplantation of the internal carotid artery to the common carotid artery, resection and shortening of the common carotid artery, common carotid artery to internal carotid artery bypass grafting, and transposition of the internal carotid artery to the external carotid artery are possible surgical techniques which are considered depending on the presence of a concomitant

atherosclerotic stenosis, length of the carotid artery, position and the complexity of the kinking, and the diameter and the quality of the adjacent arteries.^{5,6}

In our case, there was no carotid artery stenosis in the patient and the symptoms were depended on the isolated carotid kinking. Hence, we decided division and proximal reimplantation of the internal carotid artery to the common carotid artery for surgical correction of the anatomy.

In conclusion, although carotid kinks are frequently corrected during the treatment of atherosclerotic lesions of the carotid vessels worldwide as in our group, isolated carotid kinks without carotid stenosis are rarely symptomatic and require intervention. Vascular etiology should also be remarked in patients with chronic persisting neck and oropharyngeal pain and patients should be offered treatment following multidisciplinary team review.

SUPPLEMENTARY DATA

Supplementary data related to this article can be found at <https://doi.org/10.1016/j.avsg.2019.06.019>.

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