A Rare Case of Carotidynia: Aneurysm and Kinking of the Left Internal Carotid Artery

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SOUHRN

Chronická bolest v oblasti krku může mít různou etiologii včetně karotidynie, což je poměrně vzácný stav charakterizovaný bolestí v oblasti nad karotickou tepnou, u níž se předpokládá původ v postižení cév. V této kazuistice popisujeme případ 47leté pacientky s chronickou bolestí v oblasti krku, u níž byla stanovena diagnóza aneurysmatu a zalomení levé vnitřní karotické tepny. Byla provedena úspěšná chirurgická rekonstrukce bez neurologických komplikací. Tato kazuistika upozorňuje na význam časné diagnostiky a odpovídajícího chirurgického řešení přítomnosti aneurysmatu a zalomení levé vnitřní karotidy v léčbě karotidynie.

ABSTRACT

Chronic neck pain can be attributed to various etiologies, including carotidynia, a relatively rare condition characterized by pain in the region overlying the carotid artery attributed to underlying vascular pathologies. In this case report, we present the case of a 47-year-old female patient who experienced chronic neck pain and was diagnosed with left internal carotid artery aneurysm and kinking. Surgical reconstruction was performed, resulting in a successful outcome without neurological complications. This case highlights the importance of early diagnosis and appropriate surgical intervention of an aneurysm and kinking of the left internal carotid artery for the management of carotidynia.

Introduction

Chronic neck pain is a prevalent symptom encountered in clinical practice, often attributed to musculoskeletal or cervical spine pathology. However, in some cases, neck pain can be attributed to carotidynia, a rare condition characterized by pain in the carotid artery region. The etiology of carotidynia necessitates a thorough investigation of the region to rule out vascular diseases such as carotid dissection, stenosis, or occlusion, as its underlying causes and management remain poorly understood.[1]

Aneurysms and kinking of the carotid arteries are infrequent yet intriguing vascular disorders that typically manifest as parapharyngeal pulsatile masses.[2] They may exhibit partial or complete thrombosis, consequently leading to embolization or compression of neural vasculature, with additional complications including ruptures and ischemic events. Consequently, the mortality rate among non-operated patients is notably elevated.[3] The primary etiological factor implicated in this condition is atherosclerosis, with occasional instances attributed to trauma. Surgical intervention is strongly advised for symptomatic patients across all age groups.[4]

This case report presents a unique case of carotidynia in a 47-year-old female patient and describes the successful surgical intervention for the associated left internal carotid artery aneurysm and kinking, emphasizing the importance of accurate diagnosis and appropriate management in such intriguing clinical scenarios.

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Case report

A 47-year-old female patient presented with chronic, debilitating neck pain that had persisted for an extended duration. The pain was localized to the region overlying the left carotid artery. The patient described the discomfort as dull and aching, exacerbated by neck movement and palpation over the left carotid artery region. She had no associated neurological symptoms, such as weakness or sensory changes. Initial clinical evaluation revealed a tender and palpable swelling overlying the left carotid artery. The patient's medical history was unremarkable, with no history of trauma, recent infections, or inflammatory conditions. Her family history did not include any vascular disorders. Routine laboratory tests, including inflammatory markers, were within normal limits.

Given the atypical presentation of chronic neck pain and the localized tenderness over the left carotid artery, further imaging studies were pursued to investigate the underlying pathology. A Doppler USG indicated enlargement of the left carotid artery. A CT angiography was performed, which unveiled two significant findings: left internal carotid artery aneurysm measuring 2.5 cm in diameter as well as kinking and superficial pouching of the vessel (**Fig. 1**). These findings explained the patient's chronic neck pain, ultimately leading to the diagnosis of carotidynia attributed to a left internal carotid artery aneurysm and kinking.

Upon identifying the pathology, the necessity for surgical intervention was established to alleviate the patient's pain and mitigate potential complications associated with the left internal carotid artery aneurysm and kinking. The patient was informed about the risks and benefits of the treatment in detail, and upon receiving consent the patient was scheduled for surgery.

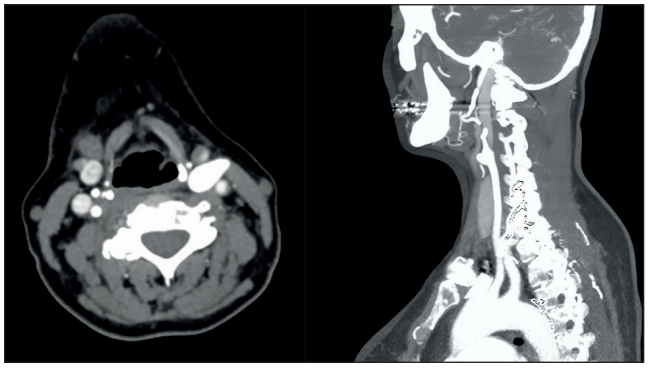

Fig. 1 – Preoperative CT angiography indicating carotid aneurysm. Note the corresponding contralateral carotid artery sizes.

Surgical management

Under regional anesthesia, the operation was initiated with a linear incision to the left cervical region anterior to the sternocleidomastoid muscle. The left facial vein was prepared for use as a patch. The left common carotid artery (CCA), internal carotid artery (ICA), and external carotid artery (ECA) were explored (**Fig. 2**), and the vessels were temporarily clamped using tape. A local anesthetic was injected into the carotid sinus to ameliorate perioperative hemodynamic instability. Following heparinization, the left ICA was clamped, and the patient's neurological functions were examined which remained stable. Following confirmation of absence in neurological and motor deficits, the CCA, and ECA were clamped as well. The aneurysmal segment of the left ICA was resected at

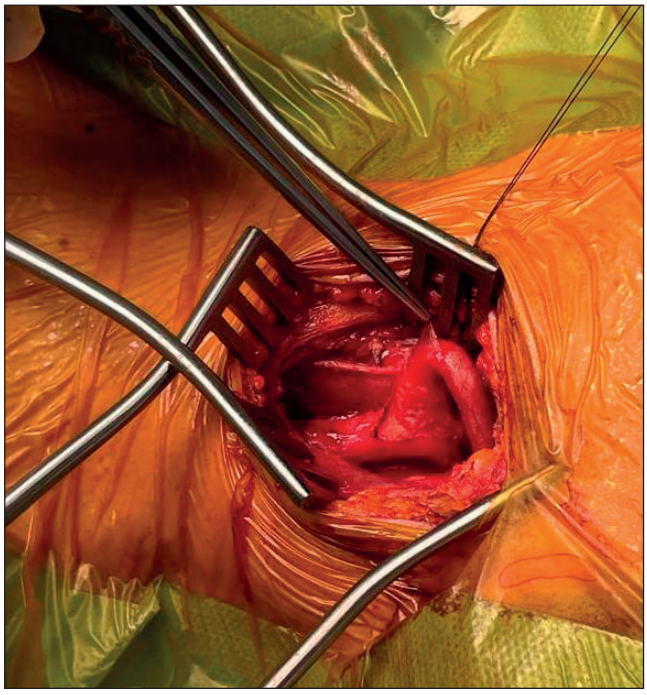

Fig. 2 – Perioperative view of the dilated and kinked carotid artery.

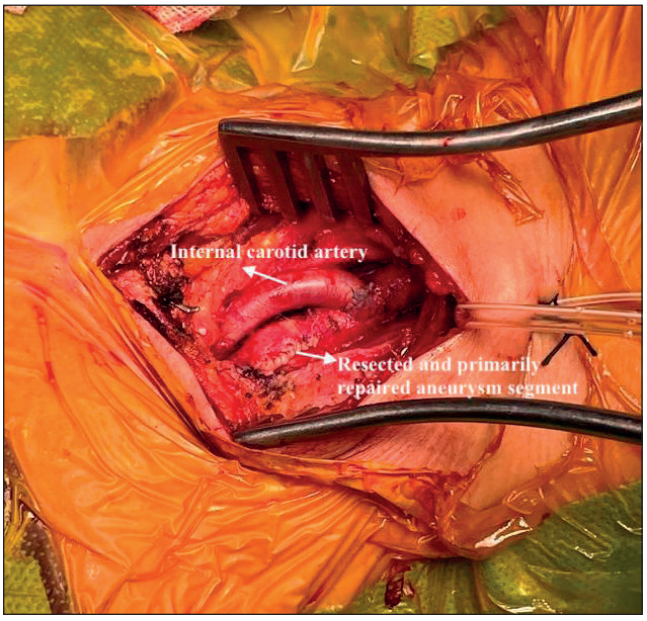

Fig. 3 – The arterial anatomy following resection of the aneurysm, reconstruction of the common carotid artery, proximal and lateral re-implantation of the internal carotid artery to the common carotid with venous patch augmentation.

the point of bifurcation. The CCA was primarily closed with Prolene sutures. An atherosclerotic segment with intimal hyperplasia was present at the orifice of the left ICA which was resected. The area where the ICA would be anastomosed to the CCA was prepared with a punch. During the end-to-side anastomosis of the left ICA to the CCA, the facial vein was employed as a patch to augment the arteriotomy (**Fig. 3**). This approach served as a prophylactic measure to prevent restenosis. The clamps were removed, and no neurological deficit was observed. The operation was concluded following appropriate surgical protocol. Postoperative imaging confirmed adequate blood flow through the reconstructed carotid artery, and the patient reported significant relief from her neck pain. The success of this surgical intervention was evident, as the patient was discharged on the second day post-surgery without any neurological deficits.

Discussion

Carotidynia, originally described by Temple Fay in 1927, is characterized by localized neck pain, often elicited by palpation over the carotid artery, without any clear signs of inflammation or infection. It remains a diagnosis of exclusion, as there are no specific diagnostic criteria, and its pathophysiology is not fully understood. The challenge in diagnosing carotidynia lies in differentiating it from more sinister conditions such as atherosclerotic disease or carotid artery dissection.[5] In our case, the diagnostic workup with a CT angiography revealed a 2.5 cm left internal carotid artery aneurysm and kinking. These imaging findings provided a clear explanation for the patient's persistent neck pain. This unique presentation highlights the importance of considering vascular etiologies in the evaluation of chronic neck pain, especially when accompanied by unusual or focal symptoms.

Kinks in the internal carotid artery can manifest as acquired or developmental anomalies. A common cause is the excessive length of the carotid artery which may result in redundancy and tortuosity of the vessel.[6] Notably, kinks are observed to be four times more prevalent in women than in men. However, the precise incidence remains uncertain due to many patients remaining asymptomatic. Symptoms associated with kinks in the internal carotid artery tend to parallel those of atherosclerotic diseases affecting the carotid artery, encompassing conditions such as transient ischemic attacks, strokes, and amaurosis.[7] The underlying pathophysiology often revolves around hemodynamic and structural abnormalities within the kinked carotid artery, leading to turbulent flow, intimal ulcerations, and subsequent embolization.[8]

Most cases of internal carotid artery aneurysms occur spontaneously, with atherosclerosis serving as the primary etiological factor, or they are a consequence of blunt trauma affecting the neck region and the high cervical segment of the internal carotid artery.[9] Blunt or nonpenetrating trauma, although more commonly associated with thrombosis of the injured vessel, can also give rise to the development of a false aneurysm. Such blunt injury to the carotid artery can lead to various abnormalities, including spasms, intimal and medial tears, dissection, and partial or complete severance of the vessel.[10] Disruption of the arterial wall's continuity emerges as the primary causal factor in the formation of an aneurysm.

Internal carotid artery aneurysms exhibit the potential to undergo partial or complete thrombosis, leading to adverse consequences such as distal embolization, compression of adjacent structures, or even rupture.[11] Consequently, patients managed conservatively face a notably elevated mortality and morbidity rate. Nevertheless, surgical intervention plays a pivotal role in averting permanent neurologic deficits arising from thromboembolism or atheroembolism. Employing advanced vascular surgical techniques, the mortality rates associated with surgical treatment are now less than 2%.

The choice of surgical technique is essential in ensuring optimal patient's outcomes. The preferred surgical treatment approach for an aneurysm of carotid arteries involves the resection of the aneurysm with subsequent restoration of blood flow. When feasible, the optimal method for addressing an aneurysmal carotid pathology is internal carotid re-implantation. Other surgical alternatives encompass resection coupled with saphenous vein interposition, internal carotid re-implantation into the common carotid artery, and extended patch angioplasty. Notably, there has been a growing utilization of endoluminal stents and stented grafts in recent times for the management of these lesions.[12]

In our case, surgical intervention was deemed necessary to address the underlying vascular abnormalities. The chosen surgical approach involved the resection of the left internal carotid artery aneurysm at the point of bifurcation, followed by the pull-down and end-to-side anastomosis of the left internal carotid artery to the common carotid artery. Additionally, patch augmentation using the facial vein was performed as a prophylactic measure to prevent restenosis. This approach aims to address both the aneurysm and the kinking of the common carotid artery, reducing the risk of complications.

While the etiology of carotidynia remains elusive, cases such as ours demonstrate the clinical significance of thorough diagnostic evaluation, particularly in atypical presentations of neck pain. This case underscores the importance of early diagnosis and appropriate surgical intervention in managing carotidynia when underlying vascular abnormalities are identified. Although our case had a successful outcome, it is crucial to recognize that carotidynia is a heterogeneous condition, and treatment strategies may vary depending on the individual patient's presentation and the underlying vascular pathology. Further research is needed to establish standardized diagnostic criteria and treatment guidelines for carotidynia.

Conclusion

This case report emphasizes the need for a comprehensive evaluation of chronic neck pain, including consideration of vascular etiologies including aneurysms and kinking. Early diagnosis and appropriate surgical intervention, as demonstrated in this case, can lead to favorable outcomes and improved quality of life for affected individuals. Further research and collaboration are essen-

tial to advance our understanding of this rare condition and refine treatment strategies for optimal patient care.

Conflict of interest
None.

Funding
None.

Ethical statement
The work was carried out in compliance with the Declaration of Helsinki.

Informed consent
The patient provided written informed consent for the publication of this case report and any accompanying images. All identifying information has been removed to protect the patient's privacy and confidentiality, in accordance with the Declaration of Helsinki.

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