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Case Report

Bilateral Internal Carotid Artery Dissection as an Uncommon Complication Following a Fall in a Marathon Runner: A Case of Eagle Syndrome

Maraton Koşucusunda Düşme Sonrası Nadir Bir Komplikasyon Olarak Bilateral İnternal Karotis Arter Diseksiyonu: Eagle Sendromu Vakası

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Keywords
Eagle syndrome
elongated styloid process
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Abstract

Eagle syndrome is a rare vascular condition characterized by the elongation of the styloid process and/or the calcification of the styloid ligament. This elongation or calcification can lead to compression or irritation of the surrounding nerves, blood vessels, and soft tissues, potentially causing compression or irritation of the extracranial part of the internal carotid artery (ICA) and resulting in symptoms such as throat pain, difficulty swallowing, ear pain, headache, and facial pain. Diagnosis of Eagle syndrome typically involves clinical assessment and imaging studies, with treatment options varying from conservative approaches to surgical interventions based on symptom severity. Patients with dissection of the extracranial part of the ICA or the vertebral artery are generally recommended antithrombotic therapy, which may include antiplatelet or anticoagulation treatment. In this case, the diagnosis and treatment of Eagle syndrome, which presented with an ischemic stroke caused by the elongated styloid process after a fall during a marathon, are discussed.

Anahtar Kelimeler
Eagle sendromu
uzamış stiloid çıkıntı
internal karotid arter diseksiyon

Özet

Eagle sendromu, stiloid çıkıntının uzaması ve/veya stiloid ligamentin kalsifikasyonu ile karakterize nadir bir vasküler durumdur. Bu uzama veya kalsifikasyon, çevredeki sinirler, kan damarları ve yumuşak dokuların sıkışmasına veya tahriş olmasına neden olabilir, bu da internal karotis arterinin (IKA) ekstrakraniyal kısmının sıkışmasına veya tahriş olmasına yol açarak boğaz ağrısı, yutma güçlüğü, kulak ağrısı, baş ağrısı ve yüz ağrısı gibi semptomlara neden olabilir. Eagle sendromu tanısı genellikle klinik değerlendirme ve görüntüleme çalışmaları ile konulur ve tedavi seçenekleri, semptomların şiddetine bağlı olarak konservatif yaklaşımlardan cerrahi müdahalelere kadar değişir. IKA'nın veya vertebral arterin ekstrakraniyal kısmının diseksiyonu olan hastalara genellikle antitrombotik tedavi önerilir, bu tedavi antiplatelet veya antikoagülan tedaviyi içerebilir. Bu olgu sunumunda, maraton sırasında düşme sonrası uzamış stiloid çıkıntının neden olduğu iskemik inme ile ortaya çıkan Eagle sendromunun tanısı ve tedavisi tartışılmıştır.

Introduction

Eagle syndrome is a rare vascular condition characterized by the contact between the elongated styloid process (ESP) and/or calcified stylohyoid ligament (CSL) with the extracranial segment of the internal carotid artery (ICA) (1). This contact can result in neurological symptoms, either through compression during rotational movements or from carotid artery dissection and/or aneurysms (2,3). The styloid process (SP) is a small, pointed bony projection located beneath the ear. In Eagle syndrome, it becomes elongated and may cause discomfort, particularly during swallowing or head movements (4,5). The etiology of ESP can be idiopathic, congenital (related to the persistence of cartilaginous elements from SP precursors), or acquired (due to osseous tissue proliferation at the stylohyoid ligament insertion) (6). This condition affects approximately 4-10% of the population. Eagle syndrome was originally classified into two types: the first being the classic styloid process syndrome, caused by fibrous tissue distorting cranial nerve endings in the tonsillar bed post-tonsillectomy; the second type results from compression of the sympathetic chain within the carotid sheath.

In this case, we will examine a case of a patient who

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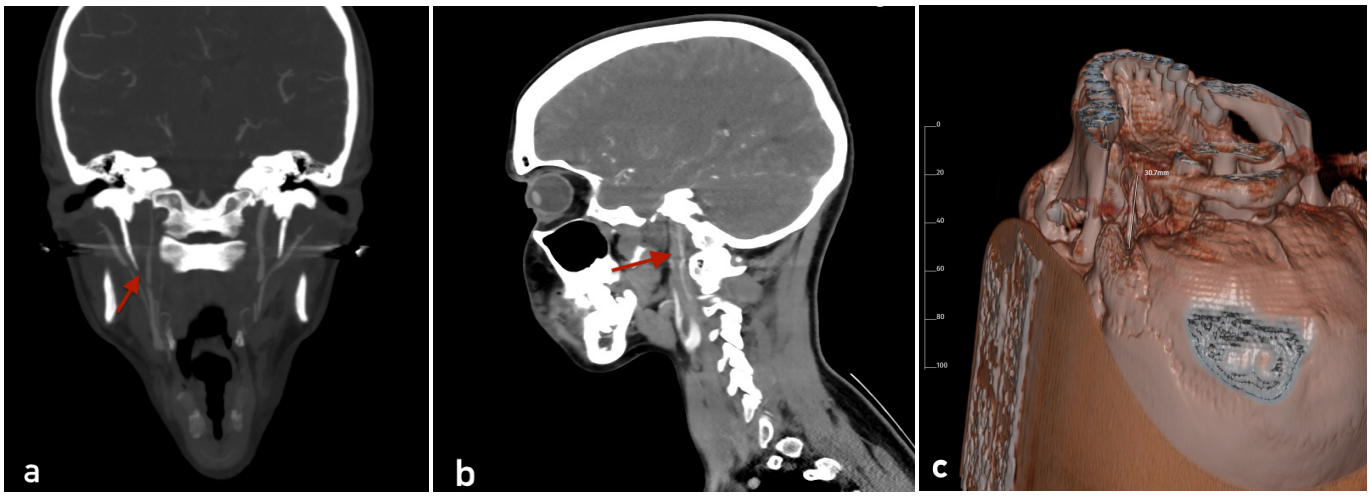

Figure 1: a) The elongated styloid process is in contact with the internal carotid artery wall, and **b)** leading to dissection of the internal carotid artery (red arrows). The volume rendering (VR) image indicates that the length of the styloid process measures 30.7 mm **(c)**.

experienced bilateral ICA dissection following a fall during a marathon.

Case Report

A 42-year-old female patient presented to the emergency department with weakness and numbness in her left arm and leg. Two weeks prior, she had a fall while running a marathon, resulting in neck pain, headache, and intermittent numbness in her left arm and leg. Upon neurological examination, she exhibited left hemiparesis and left-side hemihypoesthesia. Subsequently, the patient underwent a brain computed tomography (CT) scan due to neurological findings (Philips Ingenuity, Amsterdam, Netherlands) , which revealed to abnormalities. Following this, a Computed Tomographic Angiography (CTA) was performed, showing significant stenosis in the bilateral internal carotid arteries of the patient (Figure 1a,b). The CTA imaging in the coronal section measured the SP as 30,7 mm (Figure 1c).

A Magnetic Resonance Imaging (MRI) scan was conducted using a 1.5 Tesla (Siemens Magnetom Sola, Erlangen, Germany), which identified restriction in scattered locations in the subcortical area of the right parietal lobe, the right centrum semiovale, and in adjacent to the ventricular structures (Figure 2a-c). These neuroimaging findings confirmed acute ischemic stroke in bilateral ICA internal watershed areas. Due to patient incompatibility, Doppler ultrasonography (US) could not be performed.

A neck imaging imaging was planned a preliminary diagnosis of bilateral ICA dissection. Axial fat-suppressed T1-Weighted images showed displayed indicating indicative of hematomas observed in the cervical segments of both ICAs from the bulb onwards, suggesting dissection (Figure 3). Digital Subtraction Angiography (DSA) was not planned deemed necessary the patient patient, as the was proven confirmed through neck MRI. The patient was monitored in the neurology intensive care unit for the first initial days, then followed by an additional days more the neurology ward. Subsequently, the patient was prescribed treatment. Written consent was obtained from the patient for the presentation.

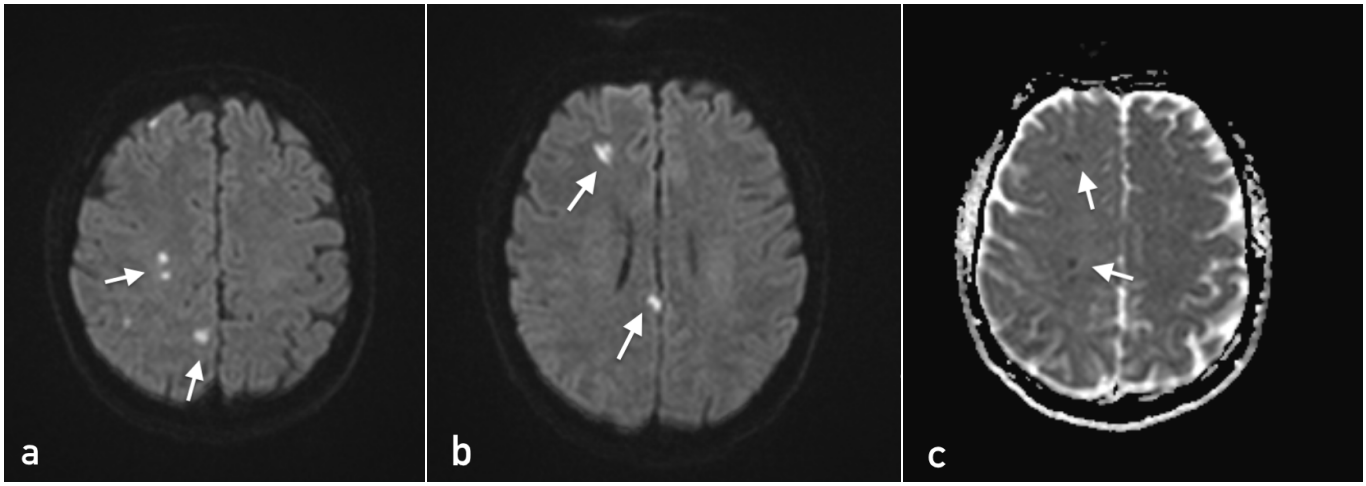

Figure 2: DWI **(a,b),** and ADC **(c)** images reveals acute diffusion restriction observed in scattered locations within the subcortical area of the right parietal lobe, right centrum semiovale, and in regions proximal to the ventricular structures (white arrows).

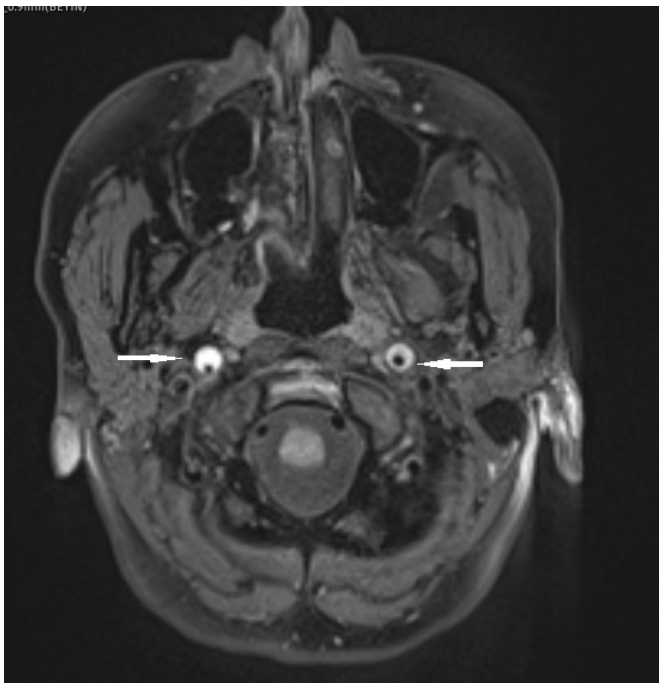

Figure 3: Fat-suppressed neck MRI reveals hyperintensities suggestive of intraluminal hematoma in the cervical segments of both internal carotid arteries (ICAs) from the bulb onwards (white arrows).

Discussion

The average length of the styloid process ranges from 15.2 to 47 mm, with lengths exceeding 30 mm potentially leading to Eagle syndrome. Rarely, patients may exhibit symptoms such as transient ischemic attacks and carotid dissection (7,8). In this particular case, the measured length of the styloid protrusion was 30 mm. Given the patient's history of sports-related trauma, it was hypothesized that there might be a dissection of the extracranial segment of the ICA based on the length of the styloid process.

Diagnosis of cervical artery dissection through imaging modalities like US, CTA, or MRI is crucial for confirmation. Preferably, brain and neck MRI is recommended, with cervical and cranial T1 fat- suppressed imaging being particularly effective in identifying small intramural hemorrhages (9). The characteristic crescent sign of intramural hematoma manifests as an eccentric hyperintense ring encircling a hypointense arterial lumen on MRI. While crescent sign is traditionally described observed in T1-Weighted fat saturation MRI sequences, this sign can also be detected in other sequences like Diffusion-Weighted Imaging (DWI) and/or CTA (3). In our specific case, US was not feasible due to patient-related constraints, leading to the performance of CTA and MRI. Consistent with existing literature, our case exhibited the crescent sign on MRI, aligning with established findings. The diagnosis was established based on typical MRI features, obviating the need for invasive procedures such as DSA.

In the management of patients with extracranial ICA or vertebral artery dissection, antithrombotic therapy, comprising antiplatelet or anticoagulation treatment, is commonly recommended. Acute anticoagulation can be initiated using subcutaneous low molecular weight heparin or intravenous unfractionated heparin. For clinically stable patients in the subacute phase, a transition to warfarin therapy is feasible, with a target international normalized ratio (INR) of 2.5 and an acceptable range of 2-3 (10).

Intravenous thrombolysis with alteplase (tPA) or tenecteplase is indicated for eligible patients with acute ischemic stroke, including those with isolated extracranial or intracranial cervical artery dissection. In cases of acute ischemic stroke resulting from extracranial internal carotid artery occlusion, early mechanical thrombectomy is recommended, especially when conducted in specialized stroke centers, irrespective of prior intravenous thrombolysis therapy (11). In the present case, as the patient's National Institutes of Health Stroke Scale (NIHSS) score was below 5, neither intravenous tPA nor thrombectomy was administered. The patient was closely monitored in the intensive care unit during the acute phase and received treatment with low molecular weight heparin. Long-term anticoagulation therapy was preferred in this instance, and following the patient's consent, treatment was arranged to maintain the INR within the range of 2-3. Notably, the patient did not encounter any complications throughout the treatment course.

Conclusion

Eagle syndrome, although rare, can have a substantial impact on the quality of life due to its persistent symptoms. The treatment primarily focuses on pain relief and functional improvement, with surgical intervention showing promising results in cases where it is deemed necessary.

Ethical Approval and Consent: Written consent was obtained from the patient for the presentation.

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