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SHORT REPORT **KISA RAPOR**

SIMULTANEOUS BILATERAL INTERNAL CAROTID ARTERY OCCLUSION:

A CASE REPORT AND REVIEW OF LITERATURE

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ABSTRACT

Bilateral ICA occlusion is a rare stroke syndrome presenting with acute onset coma with intact brainstem functions. We report herein a typical example of acute simultaneous bilateral ICA occlusion case along with a meta-analysis of all cases reported since its first description in 1954. A 83-year-old woman with known hypertension and lung cancer presented to the emergency department with sudden onset coma. On CT angiography, bilateral internal carotid artery acute occlusion was detected, and the patient was transferred immediately into the angiography suit for endovascular treatment. Recanalization couldn't be achieved, and the patient expired on second day after admission. Our meta-analysis included 25 patients from 18 different case reports. The mean age was 57±20 years, the mortality rate was 62.5% and the most common stroke risk factors were hypertension and smoking. The patients presented mostly with lateralizing hemispheric signs (56%) and imaging studies showed bilateral lesions in 72% of the cases. Based neuroimaging studies, we concluded that the mechanism of stroke was malignancy induced hypercoagulability. Acute bilateral carotid artery occlusion is a moribund condition without timely and effective recanalisation. But, difficulties in differentiation from metabolic comas and massive posterior circulation strokes often lead to delayed treatment.
Keywords: Bilateral internal carotid artery occlusion, acute onset coma, decortication rigidity, brain stem infarction, malignancy, hypercoagulability.

SİMÜLTANE BİLATERAL İNTERNAL KAROTİS ARTER OKLÜZYONU:

OLGU SUNUMU VE LİTERATÜR TARAMASI

ÖZ

Bilateral ICA oklüzyonu beyin sapı reflekslerinin korunduğu akut koma tablosu ile kendini gösteren oldukça nadir bir durumdur. Bu çalışmada ilk tanımlandığı 1954'ten itibaren bildirilen bilateral ICA oklüzyonu vakalarının meta-analizi ile birlikte ünitemizde bilateral ICA oklüzyonu ile izlediğimiz bir olguyu sunuyoruz. Bilinen hipertansiyon ve akciğer kanseri tanısı olan 83 yaşında kadın hasta acil servise ani başlangıçlı koma ile başvurdu. Beyin boyun BT anjiyografi incelemesinde bilateral karotis arter oklüzyonu olduğu tespit edilmesi üzerine ivedilikle endovasküler tedaviye alındı ancak rekanalizasyon sağlanamadı ve hasta başvurudan 2 gün sonra kaybedildi. Yaptığımız meta-analize 18 farklı yayında bildirilen toplamda 25 hasta dahil edildi. Ortalama yaş 57±20 ve mortalite %62.5 idi. Olgu popülasyonu içerisinde inme risk faktörleri arasında en sık hipertansiyon ve sigara yer almaktaydı. Hastaların en sık lateralizan hemisferik bulgular ile başvurduğu tespit edildi ve vakaların %72'sinde yapılan görüntülemelerde bilateral sulama alanlarında lezyon olduğu saptandı. Hastanın yapılan nörogörüntüleme çalışmaları göz önünde bulundurulduğunda etiyoloji olarak malignite ilişkili hiperkoagülabilite olduğu kanaatine varılmıştır. Akut bilateral karotis arter oklüzyonu acil revaskülarizasyon yapılmaz ise ölümcüldür. Akut bilateral karotis arter oklüzyonu metabolik komaları ve arka sistem enfarktlarını taklit edebileceği için akla gelmez ise endovasküler tedavi şansı yitirilebilir. Bu çalışma bilateral ICA oklüzyonu olan farklı vakalara tek bir kalemde yer vererek klinisyenlerin bu konudaki bilgi ve farkındalığını artırmayı hedeflemektedir.
Anahtar Sözcükler: Bilateral internal karotis arter oklüzyonu, akut başlangıçlı koma, dekortikasyon rijiditesi, beyin sapı enfarktı, malignite, hiperkoagülabilite.

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INTRODUCTION

Sudden onset coma with decerebrate and decorticate posturing is classically associated with brainstem lesions. In the context of cerebrovascular diseases, such a lesion is usually attributed to vertebrobasilar strokes (1). In the landmark article by C.M. Fisher, 6 cases of bilateral internal carotid artery (ICA) occlusions presenting with bilateral neurological symptoms and coma similar to basilar occlusions were described in 1954 (2). The importance of the absence of cranial nerve pathologies such as oculomotor or pupillary abnormalities in bilateral ICA infarcts was therein emphasized on the differentiation from basilar occlusion. Since then, a number of cases of bilateral ICA occlusions have been reported. We report another typical and well-documented example of simultaneous bilateral ICA occlusion along with meta-analysis of case reports published since 1954.

CASE REPORT

A 83-year-old woman with known history of hypertension (HT) and lung cancer was brought to the emergency department with sudden loss of consciousness after a short lasting generalized convulsive seizure. The neurological examination at the 55th minute of the event revealed decorticated posturing, bilateral Babinski sign and fixed and dilated pupils. The ECG showed normal sinus rhythm. The brain computed tomography (CT) imaging performed in the emergency department showed bilateral hyperdense internal carotid arteries (ICAs) and middle cerebral arteries (MCAs) (Figure 1). Subsequently performed CT angiography (CTA) revealed the total occlusion of bilateral ICAs extending from the bifurcations to their respective end branches. There was no collateral blood flow on the left carotid territory, and there was little collateral blood flow on the right side from the posterior circulatory system (Figure 2). The patient was intubated, and transferred directly into the radiology suit for endovascular treatment.

Bilateral femoral artery punctures were performed on the 2nd hour of the event. Consulting with the neuroradiology team, we decided to recanalize the left ICA first, due to its lesser collateral support. Several attempts were made using aspiration and stent retriever techniques,

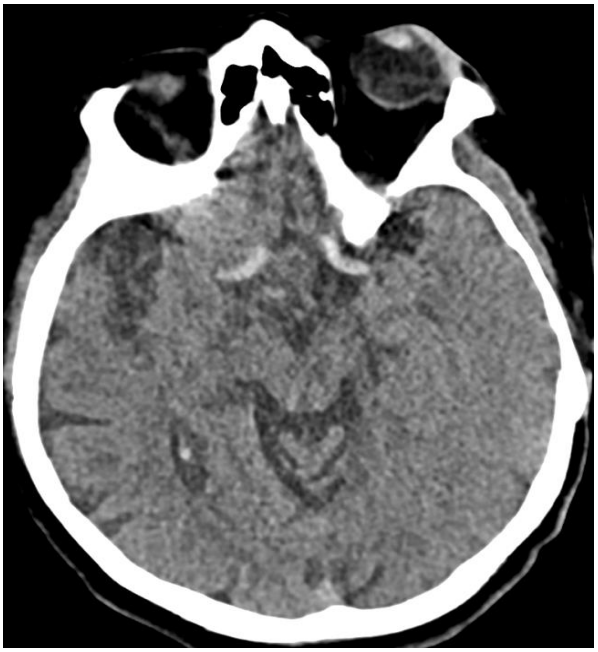

Figure 1. The CT performed at admission showing bilateral hyperdense MCAs. At first glance, it can be misinterpreted as bilateral atherosclerotic lesions.

yet the flow could not be re-established despite of a significant amount of extracted clot. After 2 hours of repeated passes without result, the neuroradiology team called off the procedure. No attempt was made for the right ICA.

The patient was then transferred to the neuro-intensive care unit (NICU). 6 hours after the procedures, a control CT was performed, revealing a hyperdense area next to the left basal ganglia, compatible with intracerebral hemorrhage. On brain MRI, diffusion weighted images (DWI) and corresponding apparent diffusion coefficient (ADC) maps demonstrated extensive acute infarctions encompassing the complete territories of MCA and ACA bilaterally, the complete territory of left posterior cerebral artery (PCA) and a small infarction located in the left cerebellar hemisphere. Moreover, T2 fluid-attenuated inversion recovery (FLAIR) sequences showed hyperintense signals consistent with vessel occlusion and/or significant slow flow through bilateral MCAs (Figure 3). A control brain CT that was performed at the 24th hour of the event displayed extensive bilateral brain edema. The patient expired 2 days after admission. Of note, informed consent was signed by the patient's relatives for this report.

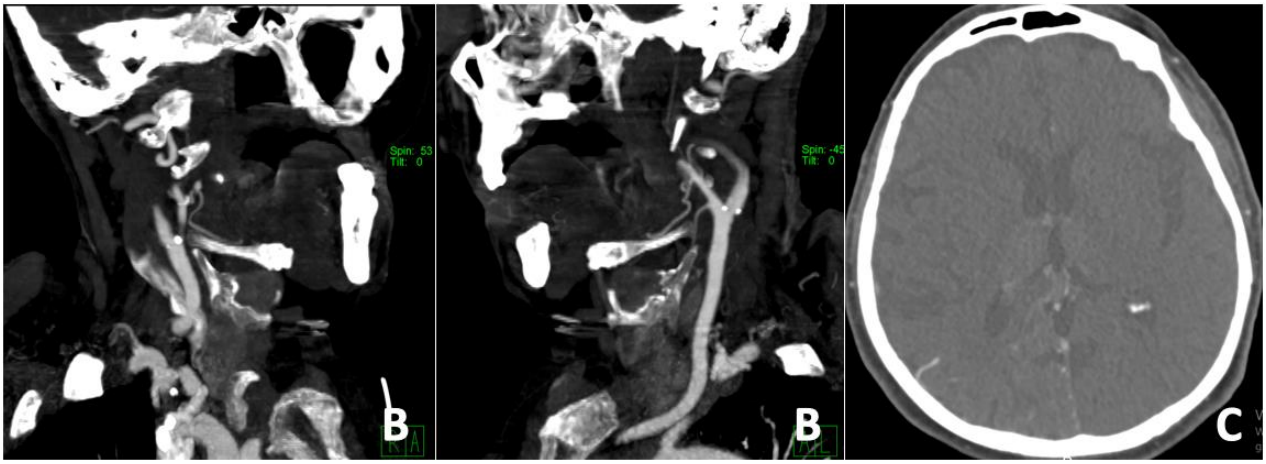

Figure 2. The CTA performed at admission. The right (A) and left (B) ICAs are occluded distally to the bifurcation level. On the axial image (C) we can see the lack of adequate collateral blood flow.

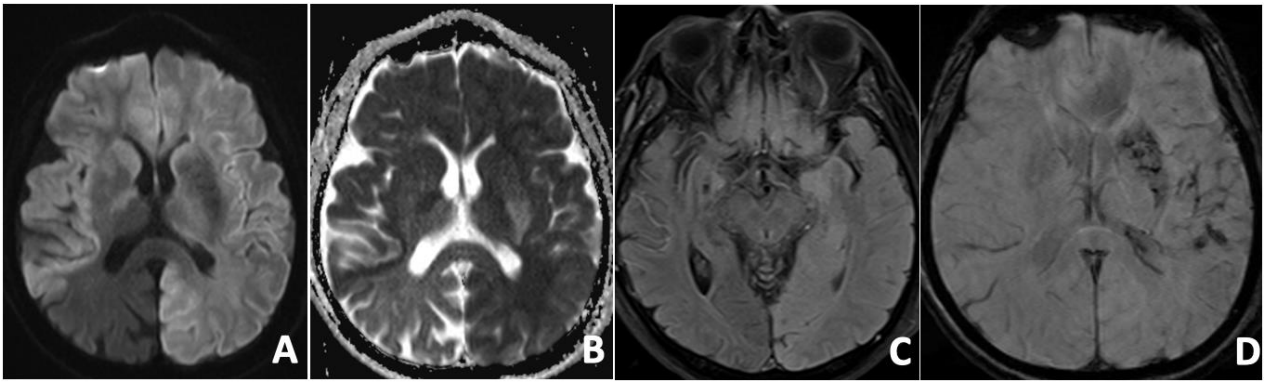

Figure 3. The brain MRI study of the case. On DWI and ADC mas, an extensive ischemic lesion encompassing the right carotid territory and the whole left hemisphere can be seen (A and B). On T2-FLAİR images, bilateral slow-flow through bilateral MCAs can be observed (C). The SWI demonstrates the hemorrhagic transformation of the left basal ganglia.

DISCUSSION AND CONCLUSION

Acute bilateral ICA occlusions generally present with acute unilateral occlusions superimposed on a contralateral chronic ICA occlusion (2-4). In our case, there was neither previous stroke history, nor any lesion on CT or MRI associated with a previous stroke episode. Furthermore, the lack of collateral blood flow on both hemispheres indicates that there was no previous chronic perfusion deficit that stimulated collateral formation. Thus, we concluded that both ICAs were occluded synchronously.

The MRI study showed us that the infarction occurred on the entire right MCA, right ACA, left MCA, left ACA and left PCA territories, and a small infarction area was present on the left cerebellar hemisphere. The DWI signals indicate that the lesions were of the same age. The CTA showed aplastic left PCA, and the fact that the left PCA territory was affected might be due to a fetal PCA pattern. The presence of the cerebellar infarction, however, indicates that the posterior circulation as also affected, possibly due to an embolic process. The ECG showed no cardiac arrhythmia, and during the patient's brief hospitalization period, no cardiac arrhythmia was detected on continuous monitoring. Thus, the etiology of the stroke was thought to be malignancy-associated hypercoagulability.

Malignancy associated hypercoagulability was first described by Trousseau in 1865, and it is considered the second leading cause of death in cancer patients (5). In the context of stroke, it is a well-known phenomenon, and it is associated with multiple vascular territory involvement (6).

Recently, a new sign called "Three Territory Sign" that is associated with malignancy induced hypercoagulability has been proposed which is also present in our case (7). Concerning bilateral ICA occlusions in the setting of malignancy, a 3-year-old male with acute myeloid leukemia (FAB AML-M0) was reported by Fuh et al. Unlike our case, the patient in this report was going through a septic course which is by itself a prothrombotic state (8).

A thorough literature search was conducted and a meta-analysis was performed in order to discuss acute bilateral ICA occlusions. The summary is given in Table 1.

The baseline characteristics are given in Table 2. Comparing our meta-analysis with the work of Wade et al. which is a prospective cohort of bilateral carotid artery occlusions (including patients with MCA trunk stenosis or occlusion, ICA stenosis or occlusion above C2 level or ICA occlusion), the patients' mean age were similar (57 ± 8 vs. 59 ± 20 years). In both cohorts, smoking and hypertension (HT) were the most frequent cerebrovascular risk factors (smoking: 85.1%, HT: 37.8% in Wade's study). Regarding the outcome, mortality was higher in our meta-analysis (62.5% vs. 11.8%) (9,10).

Clinical data regarding our meta-analysis is given in Table 3. In Wade's paper, the frequencies of involved territories are as follows: right carotid 44%, left carotid 46% and bilateral carotid 10%. They reported no vertebrobasilar involvement. Whereas in our meta-analysis, infarcts were most commonly present in both carotid territories (72%), and 3 patient (17%) had additional lesions in the vertebrobasilar circulation territory. Moreover, the most common etiologies were cardio-aortic embolism (33%) and atherothrombotic occlusions (33%). Two reports were related to hypercoagulability (one associated with malignancy and the other with Antiphospholipid Antibody Syndrome) and two reports were related to vasculitis (one associated with Behçet's Disease and the other with Giant Cell Arteritis) (8,11-13). An interesting case was that of a cannabis smoker that developed bilateral atheromatous lesion at a young age, and suffered from bilateral ICA occlusion (14).

Regarding the presenting signs, 44% of the patients presented with coma and 56% of the patients presented with lateralizing hemispheric signs. This relatively mild presentation is associated with the degree of collateral flow provided to the anterior circulation (15). Three patients presented with concomitant pupillary reflex dysfunction, but none of them had a reported midbrain infarction (one study reported that a pontine infarction could be the cause of the pupillary dysfunction) (8,16,17). Like the case in our patient, this presentation may be due to an ictal activity undetected by the investigators.

As conclusion, acute bilateral internal carotid artery occlusion is a rare condition that can mimic vertebrobasilar system infarctions. It differs from brainstem infarctions by not disrupting brainstem functions. The severity of the presenting symptoms is associated with the degree of collateral blood flow. With a high mortality rate, acute bilateral ICA occlusion has a grave prognosis.

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Table 1. Summary of the review of literature.

Year	Literature	Age, Sex	Stroke Risk Factors	Presenting Signs	Lesion Localization	Etiology	Outcome
1964	Yashon et al. (18)	25, M	None	Coma	N/A	Traumatic Dissection	Death
1978	Robinson et al. (19)	54, M	None	Lateralizing hemispheric signs	N/A	Traumatic Dissection	Death
1979	Grobovschek et al. (20)	67, M	PAD, SM, stroke		N/A	N/A	Survival with low dependency
1984	Howard et al. (13)	65, F	None	Lateralizing hemispheric signs	Bilateral carotid	Vasculitis	Death
1984	Muller et al. (21)	32, F	None	Lateralizing hemispheric signs	N/A	Traumatic Dissection	Survival with low dependency
2002	Kwon et al. (3)	58, M	SM	Coma	Bilateral carotid	Atherothrombotic	Death
		65, M	HT	Coma	Bilateral carotid	Atherothrombotic	Death
		64, M	HT, SM	Coma	Bilateral carotid	Atherothrombotic	Death
		61, F	AF	Coma	Bilateral carotid	Cardio-aortic embolism	Death
		79, F	AF	Coma	N/A	Cardio-aortic embolism	Death
		65, F	HT, AF	Coma	N/A	Cardio-aortic embolism	Death
2002	Sagduyu et al. (12)	43, M	SM	Lateralizing hemispheric signs	N/A	Vasculitis	Survival with low dependency
2003	Hagiwara et al. (16)	86, F	CHF, AF, stroke	Coma, accompanying pupillary reflex dysfunction	Bilateral carotid, accompanying posterior circulation	Cardio-aortic embolism	Death
2007	Zubkov et al. (17)	72, M	CAD, AF	Coma, accompanying pupillary reflex dysfunction	Bilateral carotid	Cardio-aortic embolism	Death
2009	Bekircan et al. (4)	91, K	AF	Coma	Bilateral carotid	Cardio-aortic embolism	Death
2010	Fuh et al. (8)	3, M	Malignancy	Pupillary reflex dysfunction	Bilateral carotid, accompanying posterior circulation	Hypercoagulability	Death
2013	Abe et al. (22)	95, F	None	Coma	Bilateral carotid	Cardio-aortic embolism	Death
2013	Tsivgoulis et al (23)	73, M	HT, HL, SM	Lateralizing hemispheric signs	Bilateral carotid	Atherothrombotic	Death
2014	Anand et al. (11)	39, F	Stroke	Lateralizing hemispheric signs	Right carotid	Hypercoagulability	N/A
2015	Xiong et al. (24)	67, M	HT, HL, SM	Lateralizing hemispheric signs	Right carotid, accompanying posterior circulation	Atherothrombotic	Survival with low dependency
2016	Jadhav et al. (25)	65, N/A	HT, HL	Lateralizing hemispheric, accompanying pupillary reflex dysfunction	Right carotid	N/A	Survival with low dependency
		55, N/A	HT, HL, SM	Lateralizing hemispheric signs	Left carotid	N/A	Survival with low dependency
		55, N/A	HT, CAD, HL, AF	Lateralizing hemispheric signs	Right carotid	N/A	Survival with low dependency
2017	Pour Rashidi et al. (26)	54, F	DM	Lateralizing hemispheric signs	Bilateral carotid	Atherothrombotic	Survival with low dependency
2019	Sharma et al. (14)	37, M	None	Lateralizing hemispheric signs	Bilateral carotid	Atherothrombotic	Survival with low dependency

M: male, F: female, SM: active smoking, HT; hypertension, PAD: peripheral artery disease, AF: atrial fibrillation, CHF: congestive heart failure, CAD: coronary artery disease, HL: hyperlipidemia, DM: diabetes mellitus; N/A: not available.

Table 2. Baseline characteristics of the meta-analysis.

Age (N=25)	59 ± 20 years
Sex (N=22)	
- Female	10 (54.5 %)
- Male	12 (45.5 %)
Cerebrovascular risk factors (N=25)	
- Hypertension	8 (32%)
- Smoking	8 (32%)
- Atrial Fibrillation	6 (24%)
- Hyperlipidemia	5 (20%)
- Previous stroke history	3 (12%)
- Coronary artery disease	2 (%8)
- Diabetes	1 (4%)
- Congestive heart failure	1 (4%)
- Peripheral artery disease	1 (4%)
- Malignancy	1 (4%)
Mortality (N=24)	15 (62.5%)

Categorical variables are presented as n (%), continuous variables are presented as mean ± SD. Due to heterogeneity of data available for each publication, each variable has its own sample universe given as N.

Table 3. Clinical features of the meta-analysis.

Presenting signs (N=25)	
- Lateralizing hemispheric signs	14 (56%)
- Coma	11 (44%)
- Concomitant pupillary reflex dysfunction	3 (12%)
Infarct localization (on CT or MRI) (N=18)	
- Right carotid territory	4 (22%)
- Left carotid territory	1 (6%)
- Bilateral carotid territories	13 (72%)
Concomitant posterior circulation territory infarction	3 (17%)
Suspected etiologies (N=21)	
- Cardio-aortic embolism	7 (33%)
- Atherothrombotic	7 (33%)
- Dissection (traumatic)	4 (14%)
- Hypercoagulability	2 (10%)
- Vasculitis	2 (10%)

Categorical variables are presented as n (%). Due to heterogeneity of data available for each publication, each variable has its own sample universe given as N.

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Ethics

Informed Consent: The authors declared that informed consent form was signed by the patient's relatives.

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