CASE REPORT

Bilateral vertebral artery occlusion after traumatic complete disruption of the cervical spine associated with ankylosing spondylitis

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ABSTRACT

Background: Bilateral vertebral artery occlusion is an uncommon and mortal injury that could be seen after high-energy traumas. We illustrate an extreme case of bilateral vertebral artery occlusion following traumatic cervical disruption with complete spinal cord injury in a patient with ankylosing spondylitis. A 49-year-old male was admitted to our emergency department after a motor vehicle accident. The American Spinal Injury Association Impairment Scale was a complete A grade. Computed tomography (CT) scan of the cervical region revealed complete disruption between C2 and C3 levels. Magnetic resonance imaging showed apparent compression and narrow calibration of the spinal cord. CT angiography demonstrated occlusion of the bilateral vertebral arteries. Because of the neurological status of the patient, extensive hematoma, and edema at the region, no surgical intervention could be planned. The patient died on the second day of his hospitalization. Only fourteen cases of bilateral vertebral artery occlusion following blunt cervical spine traumas have been reported to date. They have a possibility to cause vertebrobasilar ischemic events with a poor prognosis of morbidity and mortality. The gold standard of diagnosis is the catheter angiography, but also CT angiography has close sensitivity and specificity. The treatment strategies of vertebral artery occlusion are still unclear.

Keywords: Ankylosing spondylitis; bilateral vertebral artery occlusion; disruption of cervical spine; trauma; vertebral artery angiography.

INTRODUCTION

About 0.5% of all spinal trauma patients are associated with vertebral artery injury.[1,2] However, bilateral vertebral artery occlusion is even more rarely and could lead to severe morbidity and mortality.[3,4] Here, we report an extremely rare case of bilateral vertebral artery occlusion after traumatic disruption of the cervical spine with complete spinal cord injury in a patient with ankylosing spondylitis.

CASE REPORT

A 49-year-old male was admitted to the emergency department of our hospital after a motor vehicle accident. He was conscious and quadriplegic when he was first seen by the emergency physician. In a short span of time his neurological status was deteriorated progressively. After respiratory arrest, he was entubated swiftly. He was transferred to our intensive care unit on a trauma board. He was unconscious with a Glasgow Coma Scale Score of 3, without spontaneous breathing. The American Spinal Injury Association Impairment Scale was grade A complete. His neck was become two times thicker than normal in a short time period.

Computed tomography (CT) of the cervical region revealed complete disruption between C2 and C3 (Fig. 1a). Cranial CT was negative. Magnetic resonance imaging showed apparent compression and narrow calibration of the spinal cord, epidural hematoma of two cm, and widespread hematoma at paraspinal muscles (Fig. 1b). CT angiography of the neck

Cite this article as: Benek B, Yılmaz H, Akçay E, Yurt A. Bilateral vertebral artery occlusion after traumatic complete disruption of the cervical spine associated with ankylosing spondylitis. Ulus Travma Acil Cerrahi Derg 2021;27:697-701.

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Ulus Travma Acil Cerrahi Derg 2021;27(6):697-701 DOI: 10.14744/tjtes.2020.63679 Submitted: 06.11.2019 Accepted: 08.04.2020

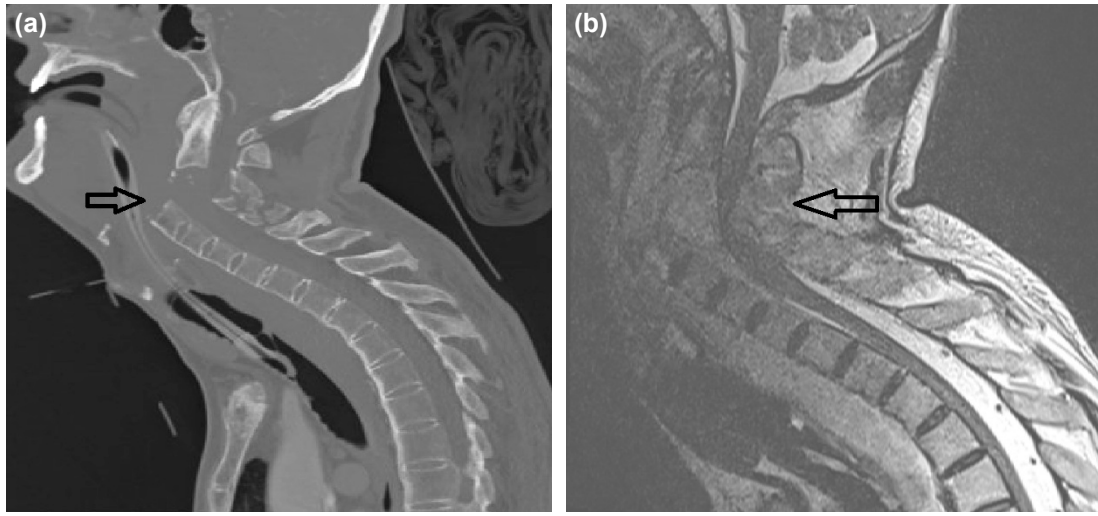

Figure 1. (a) Sagittal computed tomography scan showing complete diastasis between the vertebral bodies of C2 and C3 (black arrow). **(b)** T2 weighted sagittal magnetic resonance imaging displaying anterior cervical dislocation fracture at C2-3, apparent compression, and narrow calibration of the spinal cord (black arrow).

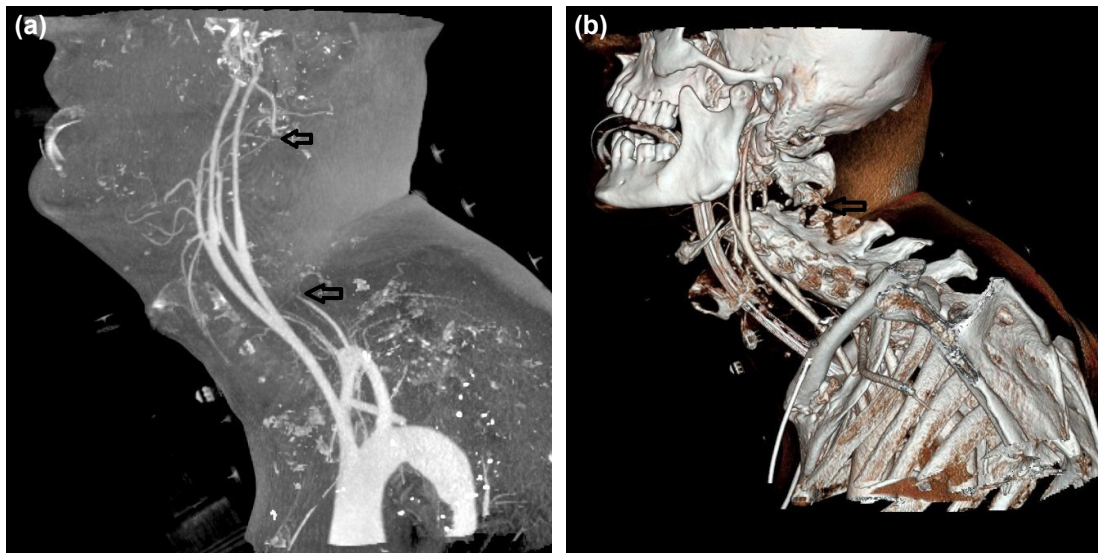

Figure 2. (a, b) Sagittal reconstruction of computed tomography (CT) and CT angiography revealed bilateral termination of the vertebral arteries and no blood flow was seen (black arrows).

demonstrated bilateral vertebral artery termination at the level of C5-6, no blood flow was seen at bilateral vertebral arteries, low stream at bilateral ICAs, retrograde flow at basilar artery, and distal vertebral artery of one or two centimeters (Fig. 2a, b). Because of the neurological status of the patient, extensive hematoma, and edema at the region, no surgical intervention could be planned. The patient died on the second day of his hospitalization.

DISCUSSION

Ankylosing spondylitis is a seronegative, chronic and progressive rheumatic spondyloarthropathy primary impinging on the spine and the sacroiliac joints.[5] It is a diffuse inflammatory disease characterized with the extensive ossification of intervertebral discs, spinal ligaments, and joints. It causes spinal bony rigidity like a bamboo spine.[6] The patients with ankylosing spondilitis are quite prone to spinal deformity and major neurological injury after cervical fractures caused by even minor traumatic forces. These kinds of traumas could lead to severe morbidity and mortality. Complete cervical disruption in a patient with ankylosing spondylitis is extremely uncommon and mortal injury.[7,8] There has been reported only one case of survival in the literature.[8]

Vertebral artery injury following blunt cervical trauma occurs in 17.2% in recent literature.[3] It is thought to be infrequent before, but today with the increasing use of angiography it is easy to be diagnosed. This vascular injury could be due to even direct trauma to the vessel wall or dissection, thrombosis, and occlusion after stretching of the intima and media of the vessels.[3,4] Vertebral artery has four segments

Table 1. Bilateral vertebral artery occlusion after cervical spine trauma-14 studies within the literature

Author	Gender/Age	Cervical fracture	Cervical treatment	Vertebral artery treatment	Outcome
Simeone and Goldberg, 1968[15]	Male/40	C3-4, C5-6	None	Open clot evacuation	Death 2 days after
Six et al., 1981[16]	Female/25	C2-3 subluxation	C1-4 posterior fusion	Anticoagulation	Cure
Kikuchi et al., 1993[17]	Male/27	C1-2 dislocation	C1-3 posterior fusion	None	Sequelae
Miyachi et al.,1994[13]	Male/49	C4-5 locked facets	C4-5 anterior fusion	None	Death 3 months after
Ozveren et al., 1999[12]	Female/41	C5-6 bilateral locked facets	C3-7 posterior fusion then C6 anterior corpectomy	Anticoagulation, Hyperperfusion	Death 3 days after
Muratsu et al., 2005[18]	Male/75	C1 anterior and posterior arch fractures	Halo then collar	Anticoagulation	Sequelae
Taneichi et al., 2005[3]	Male/34	C5 burst fracture	None	None	Sequelae
Yoshihara et al., 2011[11]	Male/67	C5-6 distraction	None	None	Death 1 day after
Jang et al., 2014[19]	Male/48	C3-4 dislocation fracture	C3-4 anterior and posterior fusion	Anticoagulation, increased fluids	Sequelae
Matsumoto et al., 2014[7]	Female/2	C5-6 complete disruption and diastasis	C5-6 posterior fusion	None	Sequelae
Golinvaux et al., 2015[9]	Male/57	C1 Jefferson fracture	Halo	Tissue plasminogen activator	Death after 2 days
Komatsu et al., 2016[10]	Male/86	C5-6 distraction	C5-6 posterior fusion	None	Sequelae
Elder et al., 2018[14]	Male/55	C5-6 complete dissociation	Halo	None	Death on first day
Strickland et al., 2019[4]	Male/53	C5-6 bilateral locked facets	C5-6 anterior fusion	None	Sequelae
Current study	Male/49	C2-3 complete disruption	None	None	Death after 2 days

anatomically: V1 (extraosseous segment), from the origin of the subclavian artery to the foramen transversarium of C6; V2 (foraminal segment), through the foramen transversarium (C6-C1); V3 (extraspinal segment), C1 to entrance into the dura; and V4 (intradural segment), from dural penetration to termination to basilar artery.[2] The second segment is the most common part for vertebral artery injuries following cervical spinal trauma.[3]

Furthermore, bilateral vertebral artery occlusion is also a rare and lethal injury. In the presence of occlusion, the collateral circulation through the circle of Willis plays an important role to preserve the function in cerebellar and brainstem structures. Ischemia symptoms often manifest in the first 24 h after trauma.[3] Bilateral vertebral artery occlusion has been only seen in fourteen cases following blunt cervical spine traumas in the literature (Table 1).[3,4,7,9–14] Several treatment modalities for vertebral artery injuries, including systemic anticoagulation, endovascular approach, surgical clot evacuation, and no treatment, were attempted before. Four of these patients died just after the days of admission,[9,11,12,14] the others lived with sequelae except one with cure. Even though Golinvaux et al.[9] had emphasized the necessity of well-defined guidelines for vertebral artery injuries, it is hard to develop it because of its rarity. Moreover, seven cases have no treatment for vascular injury and five of them were living at last follow-up.[3,4,7,10] The bilateral vertebral artery occlusion following blunt cervical trauma occurred mostly after subaxial traumas in ten cases, after upper cervical trauma in five cases including our own.[4,11]

Conclusion

Only fourteen cases of bilateral vertebral artery occlusion associated with blunt cervical spine traumas have been reported in the literature. They might cause vertebrobasilar ischemia which leads to high morbidity and mortality. The gold standard of diagnosis is catheter angiography, but also CT angiography has high sensitivity and specificity. The treatment strategies of vertebral artery occlusion are still unclear.

Informed Consent: Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

Peer-review: Internally peer-reviewed.

Authorship Contributions: Concept: B.B., H.Y., E.A., A.Y.; Design: B.B., H.Y., E.A., A.Y.; Supervision: B.B., H.Y., E.A., A.Y.; Materials: B.B., H.Y., E.A., A.Y.; Data: B.B., H.Y., E.A., A.Y.; Analysis: B.B., H.Y., E.A., A.Y.; Literature search: B.B., H.Y., E.A., A.Y.; Writing: B.B., H.Y., E.A., A.Y.; Critical revision: B.B., H.Y., E.A., A.Y.

Conflict of Interest: None declared.

Financial Disclosure: The authors declared that this study has received no financial support.

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OLGU SUNUMU - *ÖZ*

Ankilozan Spondilit olgusunda travmatik servikal komplet ayrışma kırığı sonrası iki taraflı vertebral arter oklüzyonu

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İki taraflı vertebral arter oklüzyonu yüksek enerjili travmalar sonrası görülebilen nadir ve ölümcül bir yaralanmadır. Ankilozan spondilit hastasında, travma sonrası komplet spinal kord hasarı ile birlikte servikal ayrışma kırığını takiben iki taraflı vertebral arter oklüzyonu gelişen ilginç bir olgu sunuldu. Kırk dokuz yaşında erkek hasta trafik kazası sonrası acil servise getirildi. ASIA Skalası Komplet A olarak değerlendirildi. Servikal bilgisayarlı tomografide (BT) C2 ve C3 seviyesinde komplet ayrışma kırığı saptandı. Manyetik rezonans görüntülemede spinal kordda belirgin bası ve incelme görüldü. BT anjiyografi sonrası iki taraflı vertebral arter oklüzyonu tespit edildi. Hastanın nörolojik durumu, bölgede aşırı kanama ve ödem nedeniyle cerrahi girişim planlanamadı. Hasta iki gün sonra kaybedildi. Literatürde günümüze kadar bildirilen sadece 13 servikal spinal travma sonrası iki taraflı vertebral arter oklüzyonu olgusu mevcuttur. Morbidite ve mortalite ile birlikte kötü prognoza neden olabilecek vertebrobaziler iskemi görülebilir. Tanıda altın standart katater anjiyografi olsa da BT anjiyografinin de yakın duyarlılık ve özgüllüğü vardır. Vertebral arter oklüzyonu tedavi stratejileri hala netlik kazanmamıştır.

Anahtar sözcükler: Ankilozan spondilit; iki taraflı vertebral arter oklüzyonu; servikal ayrışma kırığı; travma; vertebral arter anjiyografisi.

Ulus Travma Acil Cerrahi Derg 2021;27(6):697-701 doi: 10.14744/tjtes.2020.63679