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LETTER TO THE EDITOR

Recurrent stereotyped TIAs: atypical Bow Hunter's syndrome due to compression of non-dominant vertebral artery terminating in PICA

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Dear Editor,

Bow Hunter's syndrome (BHS) is a neurovascular condition characterized by rotational occlusion of the dominant vertebral artery (VA), causing vertebrobasilar insufficiency [1]. Symptoms appear every time dominant VA is compressed by turning the head to the opposite side; they are usually transient and reversible by turning to the neutral position [2].

A 71-year-old woman was admitted for evaluation of acute stroke with rapid onset of dizziness and impaired standing ability. On admission, neurological examination revealed mild ataxia, vertigo, horizontal diplopia, Horner's sign, peripheral facial palsy on the left, right-beating and torsional nystagmus, and lateral gaze palsy toward the left, as shown in Online Resource 1. A non-enhanced computed tomography (NECT) was unremarkable and a computed tomography angiography (CTA) of the intracranial and extracranial vessels showed that the left posterior inferior cerebellar artery (PICA) originated from the left non-dominant VA, which entered the inter-transversal canal at C5 level (Fig. 1). The patient was supposed to have acute brainstem ischemia and was treated with systemic thrombolysis with a complete recovery. Routine diagnostic workup for ischemic stroke was unremarkable. Magnetic resonance imaging (MRI) of the brain showed no infarction and evidenced a right-dominant VA (Online Resource 2).

Three months later, the patient presented with the same clinical syndrome and tinnitus without hearing loss.

NECT and CTA were unremarkable and she was treated with systemic thrombolysis again, with a complete recovery. In the following months, the patient presented four further identical episodes, all with spontaneous recovery in less than 3 h.

A year later, she had a TIA during a phone call so that we could hypothesize that rightward head rotation might be the triggering factor of the TIAs. By performing transcranial Doppler ultrasound (TCD) examination in neutral conditions and after head-turning, she again experienced vertigo, diplopia, and left facial palsy, and, in the meanwhile, we could demonstrate a reduction in blood flow in left VA (Fig. 2), thus allowing a definite diagnosis of BHS.

To better evaluate the osseous structures, a NECT of the cervical spine was performed and showed asymmetry with scoliosis with a small osteophyte before the entry of the VA into the transverse canal at C5 level.

Because of absent direct external compression cause, the patient was not a possible candidate for surgical treatment; a cervical collar avoiding prolonged head rotation was the only possible solution, which prevented further episodes in the following 2 years.

We report a patient with BHS, affecting non-dominant VA with left vertebrobasilar tract distal to PICA, who experienced 7 stereotyped and recurrent TIAs [3]. However, direct evidence of the effect of head rotation was only obtained with dynamic TCD in different head positions.

In most cases, the cause of VA occlusion is an anatomical compression by cervical vertebrae, secondary to head-turning to either or the ipsilateral or contralateral

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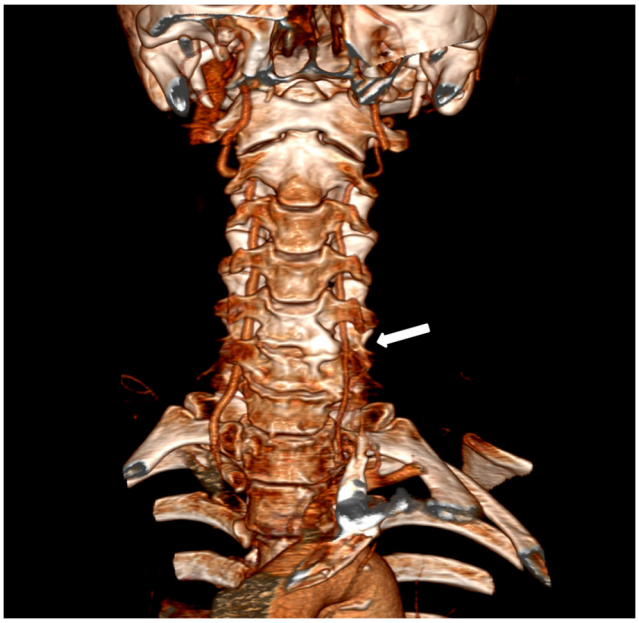

Fig. 1 Right VA entered inter-trasversal canal at C6; the arrow indicates a small osteophyte before entry of the left VA into the transverse canal at C5 level. VA vertebral artery

side. Though in several described cases, an external cause of compression of VA is found (osteophytes, bone spurs) [4], in our patient, an abnormality of entry of the left VA would explain a greater susceptibility to compression at the rotation of the head contralateral during its long path. The site of VA compression was noted to be at the level of the entrance of left VA in the inter-transversal canal at C5 level, different from most of BHS cases [2].

Furthermore, we describe an abnormal clinical presentation of BHS, which resembles a "PICA syndrome" [4].

In most patients affected by BHS, VAs usually allow for compensation in the instance of unilateral disease [2], but in our patient, compression of the left non-dominant VA evoked the symptoms, though right VA was patent. We suggest that anatomical variants highlighted in CTA play an important role to determine atypical BHS. In our patient, the right VA provides the contribution for most of the vertebrobasilar circulation, with the exception of the contralateral PICA territory. Presumably, during right head-turning, the left VA experiences a significant drop in blood flow compromising the ipsilateral PICA territory, especially in its distal branches, while the basilar artery, the contralateral PICA, and both anterior inferior cerebellar arteries were patent and fed the posterior circulation.

To our knowledge, this is the second described case of BHS affecting the non-dominant VA with exclusive PICA involvement [5]. Our patient developed nystagmus beating on the right side as in the patient described by Noh [5, 6], who had experienced isolated vertigo and nystagmus. In both cases, nystagmus had a contralateral beating direction, at the difference with nystagmus observed in Wallenberg's syndrome (ipsilateral to the lesion), but in our patient, facial nerve palsy provided evidence of the site of ischemia. We hypothesize that excitation of the vestibular nuclei, due to transient ischemia or borderzone phenomena, is the cause of the finding.

Ischemic changes of BHS are transient, though permanent deficits have been described in the case of late diagnosis [2, 7]. For this reason, BHS should be considered in patients with stereotypical and relapsing symptoms of posterior circulation ischemia, especially if related to head position.

Conclusions and limitations

We have presented a rare case of atypical BHS due to compression of non-dominant vertebral artery with anatomical variants, resulting in stereotyped and reversible PICA syndrome. BHS is a unique clinical entity that must be considered in the evaluation of patients with stereotypical and relapsing symptoms of posterior circulation ischemia. Anatomical variants can confuse the clinical picture and complicate the differential diagnosis because they resemble focal and lateralized disorders. In our patient, an anomalous level of the entrance of left VA in the inter-transversal canal and its compression during head rotation caused the symptoms.

The patient received several erroneous diagnoses before obtaining the correct one, thus experiencing up to seven identical TIAs. Due to the risk of infarction, early diagnosis of BHS is necessary.

We think that this rare syndrome should be considered in patients with stereotypical and relapsing symptoms of posterior circulation ischemia. In our case, dynamic TCD evaluation of VAs in different neck positions was a crucial tool in order to obtain a definite diagnosis, but TCD is difficult to perform in the acute setting. In fact, even if it is a cheap and low-cost technique, it requires high experience of a trained examiner in to obtain reliable results. Unfortunately, we cannot provide any colored image for technical issues.

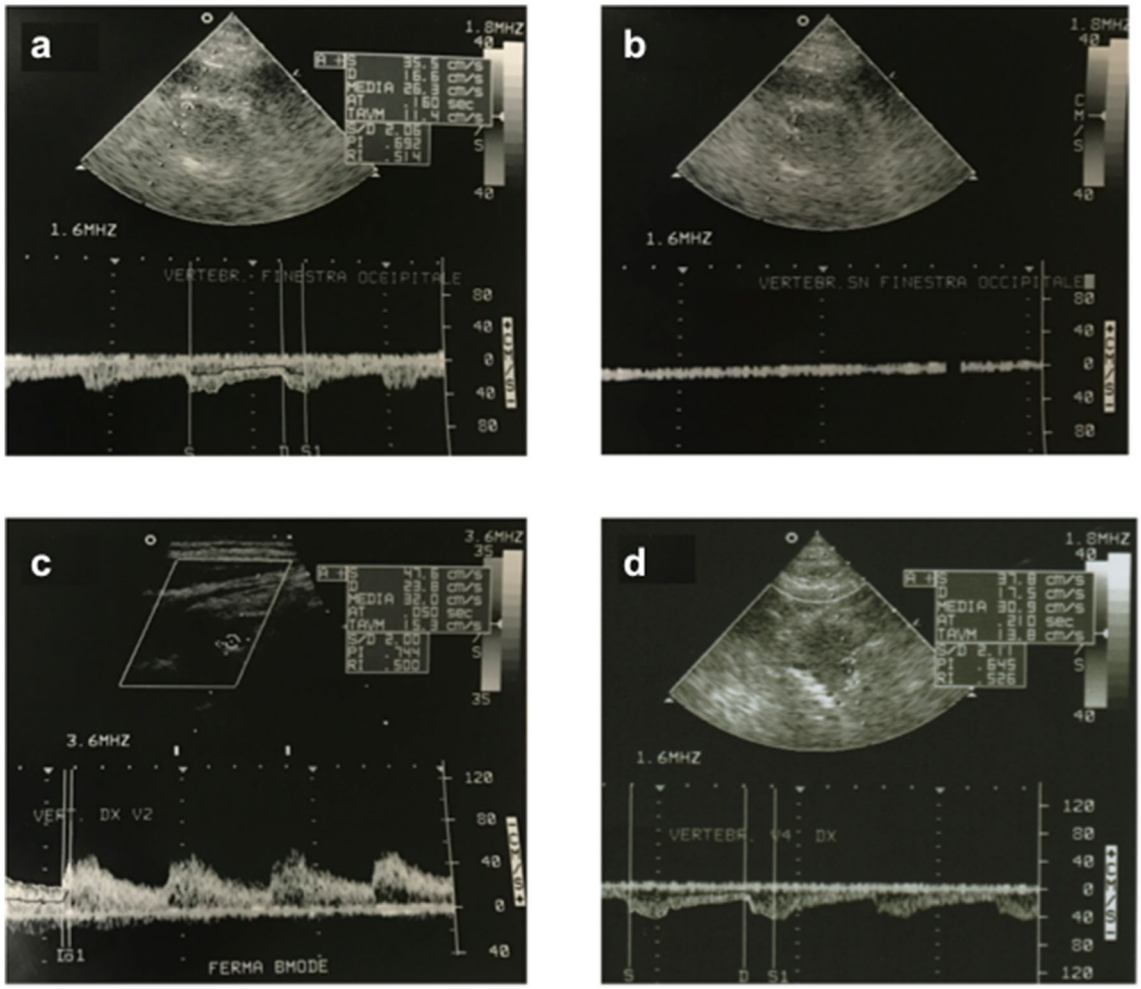

Fig. 2 TCD was performed in supine neutral position and turning the patient's head to the right. **a** Baseline TCD, occipital window: left VA (blue) with normal flow on neutral position (systolic peak velocity 35.5 cm/s; diastolic peak velocity 16.5 cm/s). **b** TCD occipital window during rightward head rotation: absence of flow in the left VA, while experiencing the symptoms. **c** Extracranial vessel Doppler ultrasound shows a right VA with normal flow during rightward rotation (systolic peak velocity 47.6 cm/s; diastolic peak velocity 23.8 cm/s). **d** TCD occipital window shows right VA (blue) with normal flow during rightward rotation (systolic peak velocity 37.8 cm/s; diastolic peak velocity 17.5 cm/s), while experiencing the symptoms. TCD transcranial Doppler ultrasound, VA vertebral artery

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Compliance with ethical standards

Ethical standards All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Conflict of interest The authors declare that they have no conflict of interest.

Informant consent Obtained.

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