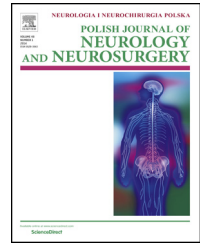


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Case report

Hypoglossal nerve palsy in the course of dissection of the internal carotid arteries – Case reports



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ABSTRACT

Internal carotid artery dissection (ICAD) has become an increasingly recognized cause of cerebrovascular accidents in young and middle-aged patients. We report 2 cases of hypoglossal nerve palsy in the course of dissection of the internal carotid arteries. The first patient was admitted to the Department of Neurology due to swallowing difficulty, speech articulation disorders and numbness of the right half of the tongue for 4 weeks. Extracranial vessel ultrasound (US) and transcranial colour Doppler (TCD) visualized thrombus causing occlusion of the right internal carotid artery (RICA). Angio-CT revealed a compression on right XII nerve and a dissection of the RICA. The second patient was referred to the Department of Neurology due to articulation disorders and swallowing difficulties. On admission, neurological examination revealed tongue deviation towards the right side with evidence of atrophy of the right half of the tongue, deviation of the uvula to the right side, absence of palatal and pharyngeal reflexes, rhinolalia and dysphagia. Vessel imaging was taken using angio-MR showing mural thrombus of the RICA.

Conclusion: The diagnosis of spontaneous non-traumatic dissection of the carotid arteries is a major challenge for clinicians. ICAD must be considered for young and middle-aged patients when severe headache is preceded by the co-existence of focal neurological symptoms. The probability of ICAD increases in the presence of predisposing diseases. The final diagnosis is based on imaging studies: color duplex ultrasound, CT angiography or MR angiography.

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1. Introduction

Internal carotid artery dissection (ICAD) has become an increasingly recognized cause of cerebrovascular accidents in young and middle-aged patients [1]. Nevertheless, ICAD remains relatively infrequent. The incidence rates are from

2.5 to 3.0 per 100,000 [2,3] and most clinicians have little experience with the condition. The variety of causative factors and clinical symptoms makes proper diagnosis difficult. Another difficulty is also related to the visualization of the dissected artery on ultrasonography and neuroimaging examinations. We report 2 cases of hypoglossal nerve palsy in the course of dissection of the internal carotid arteries.

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2. Case 1

A 50-year-old male was admitted to the Department of Neurology due to swallowing difficulty, speech articulation disorders and numbness of the right half of the tongue for 4 weeks. The patient also reported pain in the submandibular area and the right ear. The patient with suspected inflammation of the right submandibular gland was under the care of the department of laryngology. Antibiotic (amoxicillin/clavulanic acid, clarithromycin) administration did not result in improvement. On admission, physical examination revealed a homogeneous painless gland in the right submandibular area, discrete dysarthria and dysphagia, tongue deviation to the right side, a slight atrophy of the right side of the tongue, normal and symmetrical palatal and pharyngeal reflexes and asymmetry of tendon reflexes (right > left) in the upper limbs.

Laboratory tests did not reveal abnormalities (except for mixed dyslipidemia) and a routine head CT scan did not reveal any pathology. Extracranial vessel ultrasound (US) and transcranial color Doppler (TCD) visualized thrombus causing occlusion of the right internal carotid artery (RICA) in the proximal segment with compensatory acceleration of flow in the left common carotid artery (LCCA), left internal carotid artery (LICA), left anterior cerebral artery (LACA), right vertebral artery (RVA) and initiation of collateral circulation through Anterior Communicating Artery (ACoA) with the reversal of the flow direction in right anterior cerebral artery (RACA) (Fig. 1).

Angio-CT revealed a widening of the right internal carotid artery (ICA) from the division of the common carotid artery with a visible thrombus, which was 15 mm above the division completely filling the lumen of the vessel (Figs. 2 and 3). Below the carotid foramen on the basis of the skull coiling of the vessel was revealed with a clear expansion of its diameter (6–4 mm vs. 4 mm). The radiologist described compression on right XII nerve and a dissection of the RICA as the aetiology of the above symptoms.

The patient was treated with unfractionated heparin (APTT monitoring), and then the oral anticoagulation drug (warfarin)

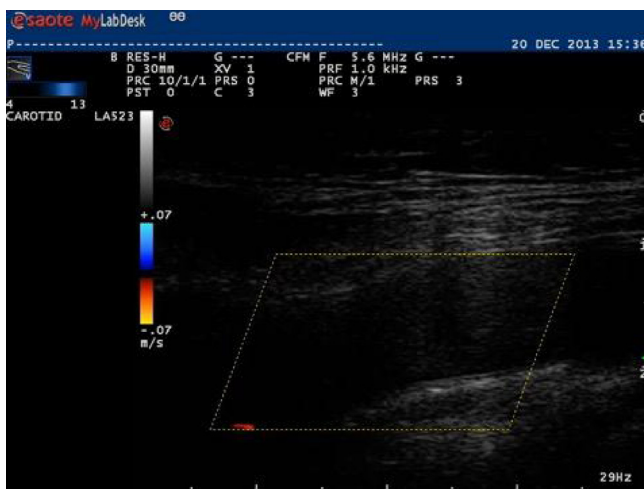


Fig. 1 – Color duplex ultrasound of carotid arteries. Case 1. Thrombus in the proximal RICA causing occlusion. Device – EsaoteMyLab60.

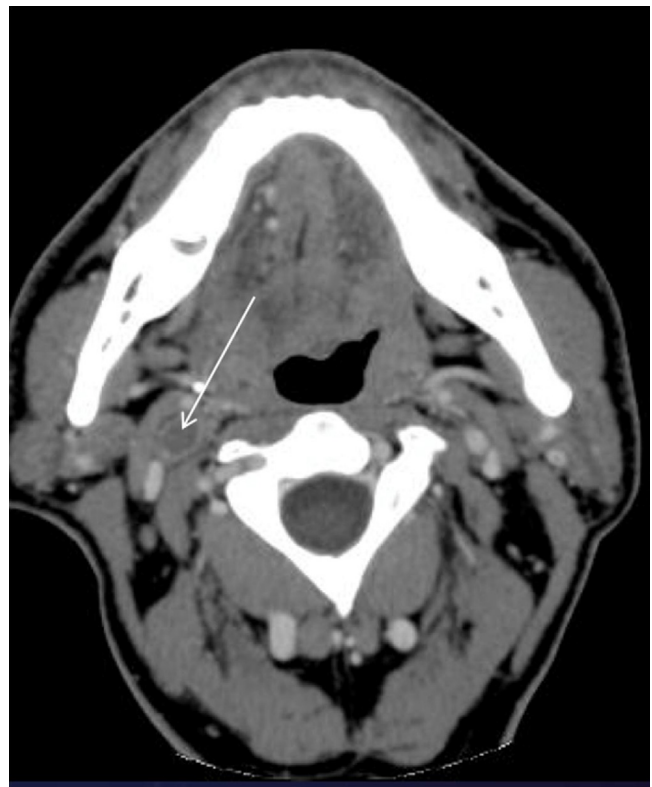


Fig. 2 – CT angiography. Case 1. Marked widening of the right internal carotid artery; about 15 mm above the division of the completely occluded vessel (no contrast enhancement). Device – CT Toshiba Aquilion 16.

was also administered. Additionally, rosuvastatin, galantamine, cefuroxime, clindamycin and lactic acid bacteria were given to the patient.

The patient was discharged with a subjective improvement in the articulation of speech and swallowing. Neurological examinations in the out-patient clinic revealed gradual regression of neurological deficits despite persistent symptoms of obstruction of the RICA on angio-CT done ~2 months after disease onset.

3. Case 2

A 48-year-old male was referred to the Department of Neurology due to articulation disorders and swallowing difficulties after a five-week treatment due to tonsillitis and sinusitis in an outpatient department of laryngology. Additionally, the patient was diagnosed with bilateral conjunctivitis and right eye keratitis. The patient's history revealed hypertension and a degenerative disease of the spine. Seven months before hospitalization the patient had participated in a car accident. At that time no injuries had been reported.

On admission, neurological examination revealed tongue deviation towards the right side with evidence of atrophy of the right half of the tongue, deviation of the uvula to the right side, absence of palatal and pharyngeal reflexes, rhinolalia (nasal speech), dysphagia (choking when swallowing liquids),

Table 1 – Comparison of tests used to diagnose dissections. Based on: Rodallec MH et al.: Craniocervical arterial dissection: spectrum of imaging findings and differentia diagnosis. Radiographics. 2008; Piątek A: Mózgowy przepływ krwi oceniony ultrasonograficznie i obrazowanie metodą rezonansu magnetycznego, a stan kliniczny pacjentów z udarem niedokrwiennym mózgu. Rozprawa doktorska 2014; Dołęga-Kozierowski B: Ocena zwężenia tętnic szyjnych wewnętrznych w angiografii konwencjonalnej 3D, dwuenergetycznej tomografii komputerowej (DSCCT) w korelacji do ultrasonografii wewnątrznaczyniowej (IVUS). Rozprawa doktorska 2014.

Examination type	Examination report summary	Benefits	Drawbacks/potential traps
Color duplex ultrasound	<ul style="list-style-type: none"> – after applying a linear transducer with a frequency range of 4–14 MHz to the patient's neck it is possible to visualize the macroscopic structure of carotid and vertebral vessels in both cross and longitudinal section – the color-coded Doppler ultrasonography enables real-time observation and archivization of blood flow characteristics, namely: blood flow direction, maximum and minimal flow velocities, flow spectrum as well as pulsatility index (PI) and resistive index (RI) 	<ul style="list-style-type: none"> – simultaneous visualization of flow spectrum and morphology of examined vessels – good availability, repeatability, non-invasive screening – it is possible to quantitatively measure the blood flow velocities and to determine the flow direction 	<ul style="list-style-type: none"> – it is not possible to visualize the whole vessel pattern – anatomical characteristics such as muscular neck, high bifurcation of the common carotid artery, calcification and iatrogenic factors constitute a significant limitation – other conditions may increase flow velocities (f.e. ICA redundancies, fibromuscular dysplasia, vasospasm, brain arteriovenous malformations, carotid cavernous fistulas, persistent trigeminal artery, anemia, and hyperthyroidism) or decrease flow velocities (f.e. occlusion of the carotid siphon, severe stenosis or occlusion of the intra-cranial ICA, and occlusion of the M1 middle cerebral artery) – the sensitivity and specificity of carotid artery stenosis evaluation is worse than in magnetic resonance and computed tomography imaging
CT angiography	<ul style="list-style-type: none"> – nonionic contrast bolus injection – scan scope: from the aortic arch to the Willis' circle (R) 	<ul style="list-style-type: none"> – accurate analysis of both lumen and wall of the vessel [r] – this method can help detect dissections and monitor vessel lesions later on [26 z Rodalleca] 	<ul style="list-style-type: none"> – slightly lower tissue resolution than in MR angiography examination – bone artifacts generated by teeth and cranial bones
MR angiography	<ul style="list-style-type: none"> – it possible to perform examination with or without contrast agent – two methods of visualizing vascular stenosis: TOF (time-of-flow) – phase contrast method with blood flow visualization, or CE-MRA (contrast enhanced magnetic resonance angiography) – where blood with contrast agent is visualized – enhancing this method with additional T1-weighted and T2-weighted sequences using fat or water saturation makes it possible to visualize the morphological structure of atherosclerotic plaques in carotid arteries – application of multi-channel imaging systems makes it possible to obtain submillimeter resolution of the examination 	<ul style="list-style-type: none"> – minimally invasive carotid arteries imaging – high sensitivity and specificity in vascular stenosis detection 	<ul style="list-style-type: none"> – absolute contraindications: presence of metal foreign bodies, devices, pacemakers, neurostimulators or medical pumps in patient's body (R) – longer examination time – lower availability
DSA	<ul style="list-style-type: none"> – generally considered the “golden standard” in carotid artery stenosis imaging, however it is not always necessary for dissection diagnosis – it is a luminographic method that makes it possible to evaluate the flow of contrast agent through the vessel 	<ul style="list-style-type: none"> – in addition it reveals intramural ulcerations (visible as excess contrast) 	<ul style="list-style-type: none"> – invasive examination – contrast agent administration – it is not possible to evaluate the vessel wall – the scope of stenosis can be evaluated only in comparison with the reference vessel section (with no atherosclerotic lesions)

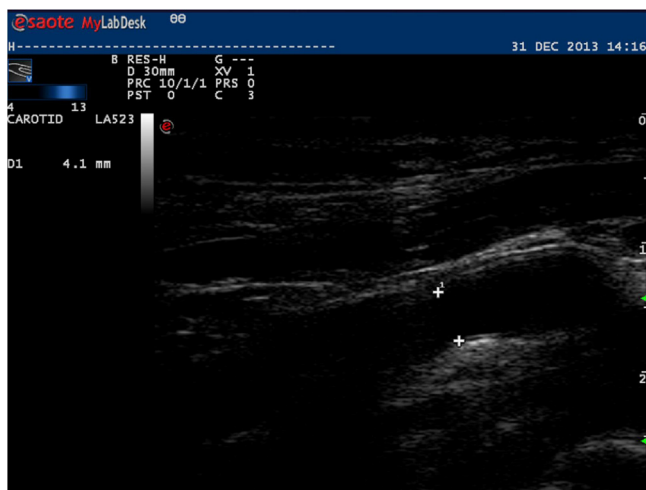


Fig. 4 – Color duplex ultrasound of carotid arteries. Case 2. Thickening of the Intima Media Thickness in the RICA. Device – EsaoteMyLab60.

manipulations [7]. Dissections can also occur in association with infectious diseases [8] and local inflammatory processes, e.g. periodontal infection [9].

The annual incidence of symptomatic spontaneous ICAD is 2.5–3.0 per 100,000 [2,3]. The incidence of carotid artery dissection as a result of blunt injuries (mainly high-speed motor vehicle accidents) ranges from less than 1% to 3% [10]. The actual incidence may be higher since some dissections are asymptomatic or cause only minor transient symptoms and remain undiagnosed. Classic manifestations of ICAD include ipsilateral pain in the neck and head, Horner syndrome and

contralateral focal cerebral lesions of ischaemic origin [11]. The neurological signs may occur almost immediately after the headache onset. This clinical manifestation is different from that usually seen in patients with extracranial ICA dissection, in which initial symptoms may precede the stroke by several days. Also, whereas patients with extracranial ICA dissection may initially experience headaches, Horner syndrome, or pulsatile tinnitus without cerebral ischaemia, patients with intracranial ICA dissection almost invariably have brain ischaemia and cerebral infarcts [12]. Fluctuation of neurological signs during the first 2 weeks after symptom onset is common, occurring in 50% of the cases. Cerebral hypoperfusion is probably the mechanism of many of these events, in contrast to distal embolism, which is thought to be the most important mechanism of cerebral ischaemia in patients with extracranial ICA dissection [12]. Cranial nerve palsy is diagnosed in 12% of adult patients with spontaneous dissection of the extracranial ICA and 5.2% patients have a syndrome of lower cranial nerve palsies (with invariable involvement of cranial nerve XII with or without additional involvement of cranial nerves XI, X, and IX) [13]. This constellation of IX, X, XI and XII cranial nerve palsies together with Horner sign is termed Villaret syndrome [14]. When the sympathetic fibres are spared – it is termed Collet-Sicard syndrome [15].

Isolated idiopathic hypoglossal nerve palsy is an uncommon cranial neuropathy, which usually presents with dysarthria, a mild tongue movement impairment and dysphagia [16]. When hypoglossal nerve palsy is not associated with other neurological deficits, clinicians should focus on lesions of the peripheral segment of the hypoglossal nerve rather than on its origin at the brainstem [17]. After leaving the medulla oblongata in the preolivary sulcus, the hypoglossal nerve passes through the hypoglossal canal. At this level metastatic tumours located at the base of the skull, traumas and

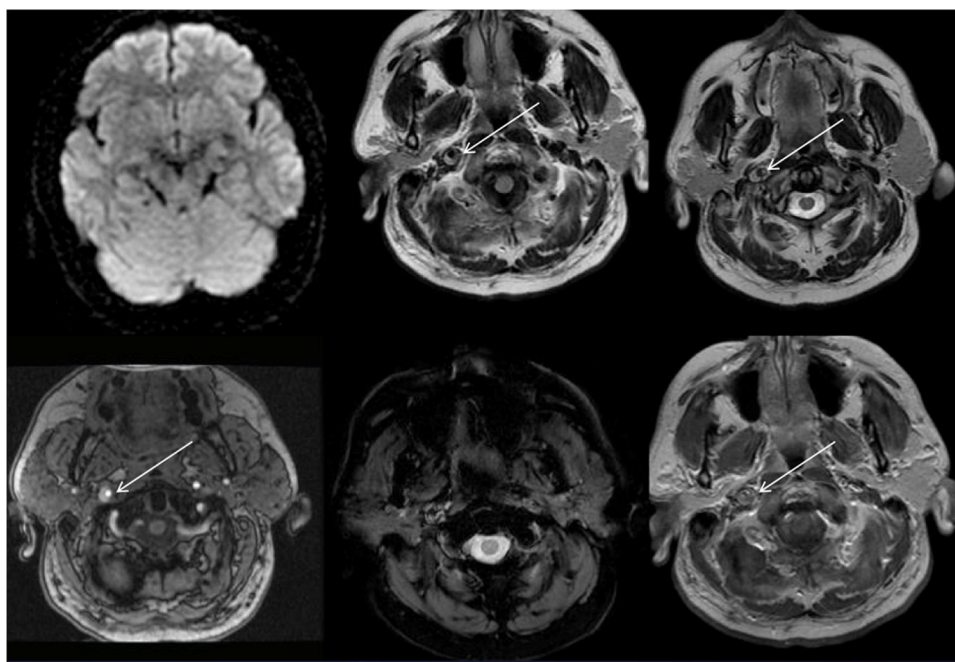


Fig. 5 – MR angiography. Case 2. Mural thrombus with widening of the lumen of the right internal carotid artery. Device – MR GE Signa HD 1,5.

inflammatory disorders are the most frequent conditions associated with palsy. Next cranial nerve XII passes between the ICA and the internal jugular vein and for this reason carotid disorders may cause nerve palsy. The hypoglossal nerve then directs towards the retropharyngeal region to finally reach the tongue. Thus pharyngeal abscesses may result in nerve palsy at this level [18].

Tests commonly used to diagnose dissections:

- Color duplex ultrasound
- CT angiography
- MR angiography
- Digital subtraction angiography

Color duplex ultrasound allows imaging of the proximal wall of ICA and visualization of blood flow velocities. This noninvasive method is quick, offers a dynamic view of vessels and does not require contrast administration. The sensitivity of color Doppler US is 95–96% for the diagnosis of internal carotid dissection causing carotid territory ischaemia and 71% for dissection causing no ischaemic events [19]. The wall of the proximal part of the ICA can be depicted with B-mode imaging and high-frequency linear transducers (4–8 MHz). Mural haematoma and thrombus may be detected as a thickened hypoechoic vessel wall. Usually, wall haematoma and intraluminal thrombus cannot be differentiated with B-mode imaging. Sometimes, an inner intimal echo helps in distinguishing wall haematoma from intraluminal thrombus or plaque in patients with thickening of the ICA wall [20]. Clinicians should be careful to diagnose ICA dissection based on US, bearing also in mind other conditions associated with increased or decreased flow velocities in the cervical ICA or occlusion of the cervical ICA [21]. Increased flow velocities may be observed in ICA redundancies, fibromuscular dysplasia, vasospasm, brain arteriovenous malformations, carotid cavernous fistulas, persistent trigeminal artery, anaemia, or hyperthyroidism [21]. Occlusion of the carotid siphon, severe stenosis or occlusion of the intra-cranial ICA, and occlusion of the M1 middle cerebral artery often show decreased flow velocities in the ipsilateral cervical ICA [21]. Our 2 cases demonstrate a variable sensitivity of US in the diagnosis of ICAD. The first case demonstrated the complete occlusion of RICA, and US findings: (1) diminished frequencies, increased resistive index, and more than a 50% reduction of flow volume in the right common carotid artery as compared with the opposite side; (2) diminished frequencies to loss of the diastolic phase, more than a 50% reduction of the flow volume, and reversed ICA/external carotid artery flow volume in the ICA as compared with the opposite side; and (3) a forward Doppler ophthalmic artery flow. The second case revealed no significant pathology in US.

The typical target picture – a narrow eccentric lumen surrounded by crescent-shaped mural thickening and thin annular enhancement is a very specific sensitive sign of arterial dissection on CT Angiography [22]. In most cases, extracranial carotid artery dissection spares the carotid bifurcation as well as the intracranial part of the ICA [3]. Most often, ICA dissection is characterized by a narrow eccentric lumen with the increase of the external diameter of the artery [22].

MR angiography (MRA) allows imaging of intravascular blood flow. Information on the blood vessel wall is obtained indirectly in the form of blood flowing or moving. MRA was the method that was used in our case 2 instead of CT angiography – it can be used in situations where ultrasonography is unexplained, results are ambiguous or clinical uncertainties exist. MRA imaging revealed perfect sensitivity and specificity of 84% and 99%, respectively, in the diagnosis of internal carotid dissection. The pseudoenlargement of the lumen in subacute dissection corresponds to the flow associated with the intramural haematoma simulating blood flow [23].

Digital subtraction angiography is not always definitive in the diagnosis of dissection because the thickness and configuration of the arterial wall are not appreciable. Nevertheless, it has been commonly regarded as the gold standard diagnostic procedure. The string sign, the angiographic hallmark of ICA dissection, is a long tapered, usually eccentric and irregular stenosis that begins distal to the carotid bulb [24]. Focal narrowing with a distal site of dilatation is referred to as the “string and pearl” sign [24].

The diagnosis of spontaneous non-traumatic dissection of the carotid arteries is a major challenge for clinicians. The analysis of our cases provides some important points. Internal carotid artery dissection must be considered for young and middle-aged patients when severe headache is preceded by the co-existence of focal neurological symptoms. The probability of ICAD increases in the presence of predisposing diseases. The final diagnosis is based on imaging studies. Ultrasound is a screening test only, and in the case of any doubt angio-MR, DSA and CT angiography should be done.

Conflict of interest

None declared.

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