SURGICAL TREATMENT IN SYMPTOMATIC STENOSIS OF THE CAROTID ARTERY AND PERSISTENT PRIMITIVE HYPOGLOSSAL ARTERY

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Persistent primitive hypoglossal artery is a rare variant of intrauterine anastomosis between the carotid and basilar arteries, which may remain in adults. The presence of this artery in carotid artery atherosclerosis increases the risks for stroke in the carotid and basilar basins. Our clinical case illustrates successful carotid endarterectomy in the presence of an ipsilateral persistent primitive hypoglossal artery under cerebral oximetry control.

Key words: *stroke, carotid artery stenosis, persistent primitive hypoglossal artery, carotid endarterectomy, cerebral oximetry.*

INTRODUCTION

Persistent primitive hypoglossal artery (PPHA) which is a persistent carotid-basilar anastomosis is a rare vascular anomaly [1]. The prevalence of the PPHA has been reported as 0.027 to 0.26% [2]. In patients with carotid artery atherosclerosis, the presence of PPHA may lead to stroke in the carotid and basilar basins [3]. Variants of treatment for an atherosclerotic lesion of the ICA with the presence of an ipsilateral PPHA are carotid artery stenting (CAS) or eversion carotid endarterectomy (ECEA). Different variants of cerebral protection have been described in these methods of treatment [3, 4]. Herein we present a case of successful ECEA in the presence of a PPHA in a symptomatic patient.

Case report

A 60-year-old woman was admitted to the Department of Neurosurgery with complaints of double vision and dizziness. Her case history revealed that in 2004 she had had an episode of weakness in the left side of the body, and in June 2019 a repeat episode with left-sided hemiparesis and diplopia in the horizontal plane.

Duplex scanning at the prehospital stage demonstrated stenosis of the common carotid artery (CCA) of more than 80%, hypoplasia of vertebral arteries. Magnetic resonance imaging of the brain showed cystic-glial alterations in the occipital lobe on the right, consequences of endured brain infarction in the basin of the right middle cerebral artery. Duplex scanning at admission confirmed stenosis of the CCA on the right of more than 80%, additionally demonstrating stenosis in the ICA ostium up to 30–40% and an abnormal vessel originating from the ICA (Fig. 1).

MR and CT angiography confirmed right CCA stenosis in the middle third of more than 80%, stenosis in the area of the bulbus of the ICA amounted to 50%, hypoplasia of both vertebral arteries, revealing PPHA on both sides, PPHA hypoplasia on the left (Fig. 2).

The circle of Willis was disconnected, with no posterior communicating arteries. Figure 3 shows a 3-D pathology model, performed in our Institute.

Based on the obtained findings, the following diagnosis was made: atherosclerosis, right CCA stenosis, hypoplasia of vertebral arteries, PPHA hypoplasia on the left, endured stroke in the basin of the right middle artery with left-sided hemiparesis in 2004, transitory ischaemic attack (TIA) in the basin of the posterior cerebral artery in June 2019, grade

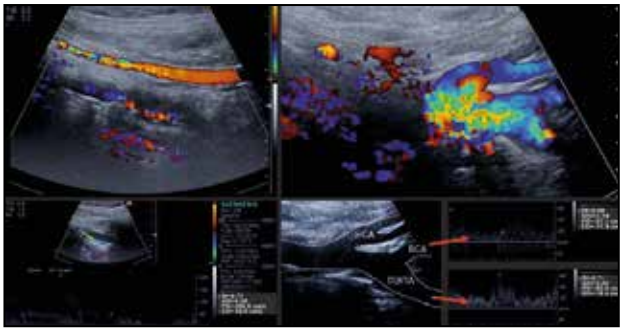

Fig. 1. Ultrasound duplex scanning. More than 80% stenosis of the CCA on the right, up to 30–40% stenosis in the ICA ostium, and an abnormal vessel arising from the ICA, with a similar curve on Doppler imaging

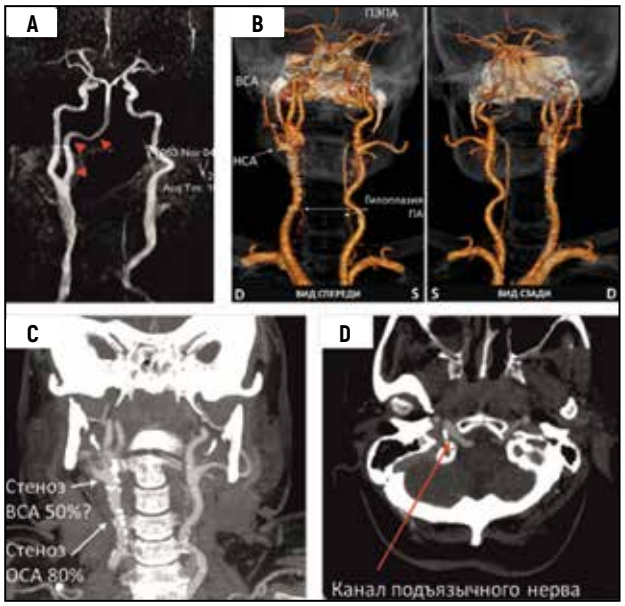

Fig. 2. Magnetic-resonance image (A) and computer tomogram (B-D), revealing hypoplasia of both vertebral arteries, more than 80% stenosis of the right CCA in the middle third, 50% stenosis in the ICA bulbus, PPHA on both sides, hypoplasia on the left

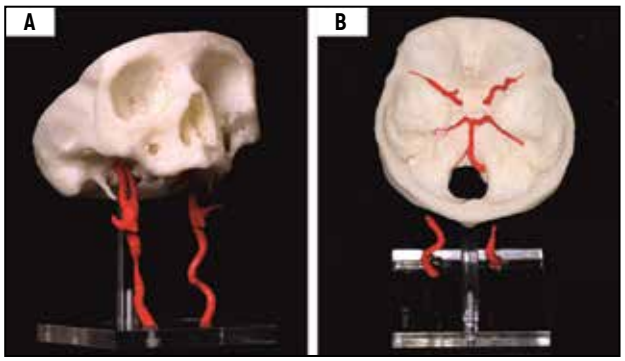

Fig. 3. 3-D pathology model: A – front view; B – back view. STL-model was performed using the program Philips IntelliSpace Portal, printed on the 3D Systems CubeX.

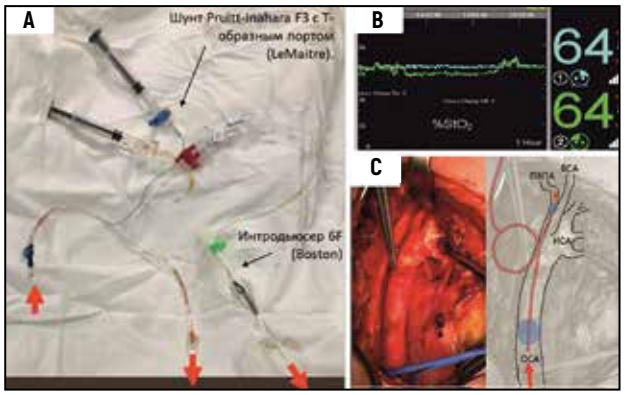

Fig. 4. Modified Pruitt-Inahara F3 shunt with a T-port (LeMaitre) supplemented with a 6F introducer (Boston) for perfusion of the PPHA and ICA simultaneously (A). The findings of cerebral oximetry on carotid artery cross-clamping showed an inconsiderable decrease in StO2 (B). Bypass grafting of only the PPHA was performed (C)

IV chronic cerebrovascular insufficiency. Given the obtained findings suggesting haemodynamically significant stenosis of the right CCA, as well as a past history of acute cerebral ischaemia (ACI) in the carotid and basilar basins, operative treatment was recommended. Given the presence of PPHA and associated risk for ACI in the basin of the basilar artery, a decision was made to perform eversion carotid endarterectomy on the right with obligatory bypass grafting of the PPHA.

Exposure revealed a high position of the carotid artery bifurcation, abnormally dilated external carotid artery (ECA). The ostia of the ICA and PPHA were exposed. Taking into account the risk for ischaemia in the carotid and basilar basins on cross-clamping of the carotid artery, we modified the Pruitt-Inahara F3 shunt with a T-port (LeMaitre) supplementing it with a 6F introducer (Boston) for perfusion of the PPHA and ICA simultaneously (Fig. 4). The findings of cerebral oximetry (cerebral tissue oximeter Fore-Sight) on carotid artery cross-clamping demonstrated an inconsiderable decrease in the StO2 on the side of cross-clamping by 11%, a decision was made to perfuse only the PPHA.

Once blood flow started along the shunt, the data of cerebral oximetry did not change. This indirectly confirmed that blood flow was filling namely the PPHA. The CCA was dissected transversely 2 cm proximal to the bifurcation. In the lumen there was a calcified

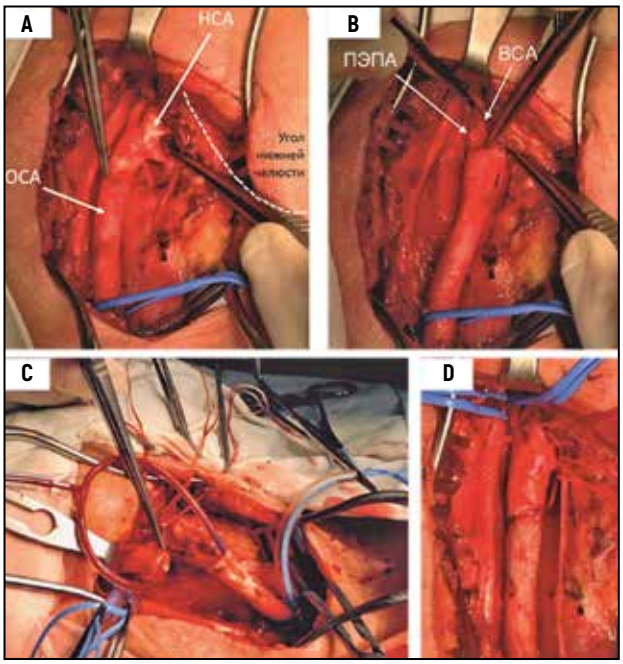

Fig. 5. Intraoperative images: A – high position of the CCA bifurcation, abnormally dilated ECA; B – separately exposed ICA and PPHA; C – CCA dissected, temporary intraluminal shunt inserted in the PPHA, retrograde eversion endarterectomy from the CCA performed, with an end-to-end anastomosis applied. D – final view of reconstruction

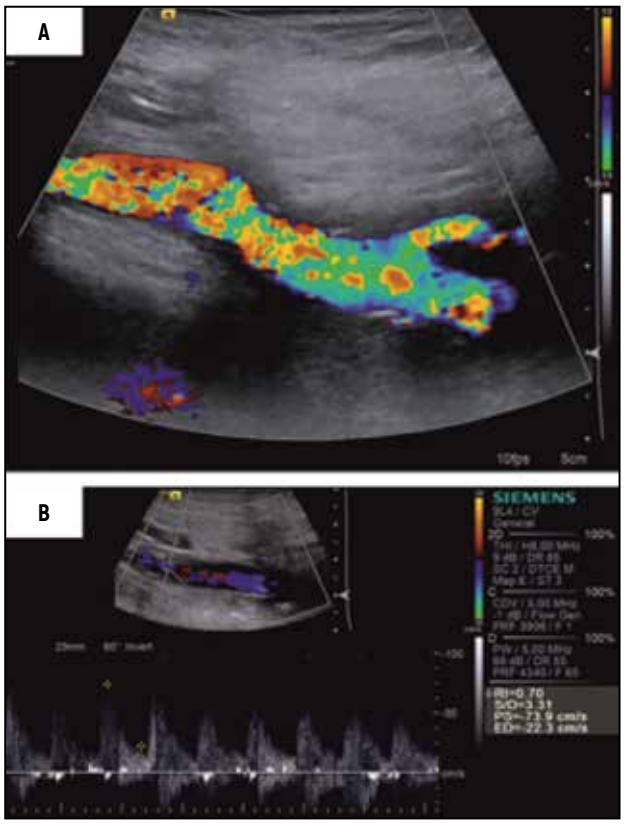

Fig. 6. Control ultrasound at 1 month after surgery. The reconstruction zone is free from restenosis signs (A) with normal velocity characteristics according to Doppler ultrasonography (B)

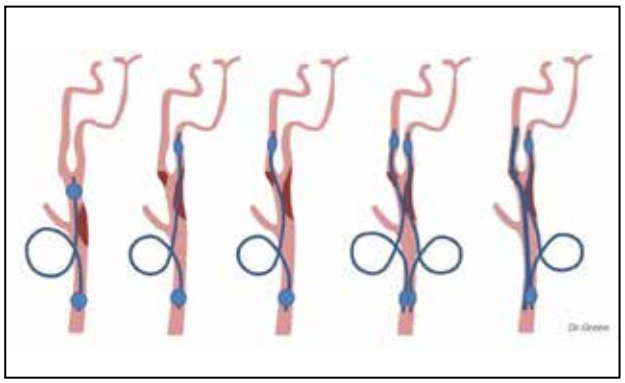

Fig. 7. Different variants of temporary intraluminal shunting

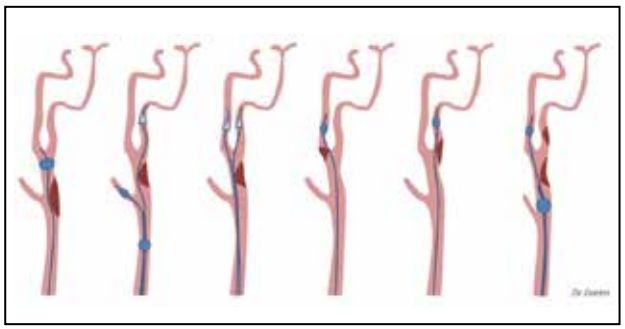

Fig. 8. Different variants of cerebral embolic protection

plaque with more than 80% stenosis. The plaque in the carotid artery bifurcation was fibrous, stenosing the lumen by not more than 30%. We performed retrograde eversion endarterectomy from the CCA 3 cm in length, with an end-to-end anastomosis (Fig. 5).

In the postoperative period, the woman showed indirect signs of cerebral hyperperfusion: pronounced headache and nausea due to high arterial pressure. On the background of correction of antihypertensive therapy her complaints disappeared. The woman was discharged in a satisfactory condition. Control ultrasonography at 1 month demonstrated no signs of restenosis in the reconstructed zone (Fig. 6).

DISCUSSION

During early embryological development, carotid–basilar anastomoses serve to perfuse the posterior circulation while the vertebrobasilar system develops and matures. These vessels involute with the emergence of the posterior communicating arteries, usually by the 40th day of fetal development [5, 6]. Preservation of this type of anastomosis sequentially leads to hypoplasia of vertebral arteries [7]. Carotid-basilar anastomoses include trigeminal, auricular, hypoglossal and proatlatal arteries. The PPHA is the second most frequently seen anastomosis after the persistent trigeminal artery [8]. It originates from C1–C2 segments of the ICA, through the canal of the hypoglossal nerve enters the cranial cavity and joins the basilar artery in the cerebellopontine angle [8, 9]. In rare cases, the PPHA originates from the ECA [10]. Bilateral PPHA was described only in 7 cases [11]. Thus, the literature presents three diagnostic criteria for PPHA: 1) artery originates from cervical segment of the ICA (type 1) or ECA (type 2) at the level of C1–C2 vertebra; 2) together with the hypoglossal nerve the PPHA passes through the canal of the hypoglossal nerve to the posterior cranial fossa; 3) the basilar artery originates from the PPHA [5]. In the majority of cases there are no posterior communicating arteries, with hypoplasia or aplasia of the ipsilateral vertebral arteries, however, this criterion was removed by J. Brismar in 1976 [2].

Although most commonly the PPHA is an accidental finding, clinical manifestations may be paresis of the hypoglossal nerve and neuralgia of the_glossopharyngeal nerve. The PPHA is often combined with various vascular pathology: aneurysms of vertebral, basilar or posterior cerebral arteries; moyamoya disease; dissection and atherosclerotic damage of the carotid artery [6].

Localization of an atherosclerotic plaque proximal to the origin of the PPHA is associated with increased risk of ACI in the anterior and posterior portions of the brain. It is necessary to exclude the presence of PPHA in development of stroke in both basins. A combination of the PPHA and an atherosclerotic lesion is a difficult

and potentially dangerous condition given the concomitant underdevelopment of carotid-basilar anastomoses and vertebral arteries. Appropriate therapeutic decision-making should begin with thorough diagnosis. Thus, in our case, duplex scanning performed at the prehospital stage had revealed no persistent anastomosis.

In their article, R. Kanazawa and coauthors presented a table including several clinical cases of persistent carotid-basilar anastomosis [12]. We supplemented this table with omitted and published later data. Surgical treatment for combined carotid artery stenosis and PPHA was described from 1978 to 2019 in 24 cases: ECEA – 16, CAS – 8 (Table) [12–21].

Table

Published cases of surgical treatment in a combination of stenosis of the ipsilateral ICA and PPHA

Author, year	Age/ gender	Side	Symptoms	Treatment	Vertebral artery	PCA
Stern, 1978 [12]*	57/f	R	Dizziness	ECEA	N. d.	absent
Pinkerton, 1980 [12]*	61/m	L	ACI	ECEA	PISA	N. d.
Osgood, 1983 [13]	57/m	R	Amaurosis	ECEA	R: N. d. L: hypoplasia	N. d.
Rodan, 1985 [12]*	41/f	L	TIA	ECEA	aplasia	absent
Ouriel, 1988 [12]*	N.d.	N.d.	TIA	ECEA	hypoplasia	absent
McCartney, 1989 [14]*	76/f	R	Amaurosis	ECEA	aplasia	N. d.
Sunada, 1991 [12]*	62/m	R	ACI	ECEA	hypoplasia	N. d.
Fantini, 1994 [12]*	67/m	L	ACI	ECEA	R: N. d. L: aplasia	N. d.
	62/f	L	ACI	ECEA	R: hypoplasia L: aplasia	absent
Cartier, 1995 [12]*	74/f	R	None	ECEA	R: aplasia L: hypoplasia	absent
Megyesi, 1997 [14]	72/m	R	TIA	ECEA	N. d.	absent
Hatayama, 1999 [12]*	71/f	L	Dizziness	ECEA	R: PISA L: aplasia	absent
Katoh, 1999[12]*	42/f	L	Fainting	ECEA	hypoplasia	absent
Bertoletti, 2000 [12]*	72/f	R	None	ECEA	N. d.	N. d.
Thayer, 2005[15]	55/f	R	None	ECEA	R: hypoplasia L: aplasia	present
Kanazawa, 2008 [12]	68/m	L	Fainting	CAS	aplasia	absent
Kawabori, 2009 [16]	71/m	R	ACI	ECEA	hypoplasia	N. d.
Nii, 2010 [3]	62/m	R	TIA	CAS	R: aplasia L: hypoplasia	absent
Silva, 2013 [17]	63/f	R	ACI	CAS	hypoplasia	N. d.
Eller, 2013 [18]	60/m	R	ACI	CAS	R: aplasia L: hypoplasia	N. d.
Huang, 2016 [19]	50/f	R	TIA	CAS	PISA	absent
Murai, 2016 [20]	77/m	R	ACI	CAS	R: aplasia L: hypoplasia	R: absent
Zhang, 2016 [4]	47/m	R	None	CAS	hypoplasia	absent
Ryu, 2016 [21]	60/f	L	None	CAS	R: hypoplasia L: narrowed	R: absent L: present

*Note: * – reference to the original source in the table presented in the article by R. Kanazawa et al. [12]; N. d. – no data; ACI – acute cerebral ischaemia; TIA – transitory ischaemic attack; ECEA – eversion carotid endarterectomy; CAS – carotid artery stenting; PISA – posterior inferior cerebellar artery; PCA – posterior communicating artery; R – right; L – left.*

The median age was 63 years, with a 1:1 male-to-female ratio. In the majority of cases, the operation was performed on the right (65%; 15/23; in 1 case no data available). Symptomatic damage (TIA, ACI, amaurosis) was observed in 62% of cases (15/24). Till 2008, only ECEA was had been performed, later on in the majority of cases – CAS. With a high level of the PPHA origin and prolonged atherosclerotic lesion, preference is given to CAS due to better visualization of the lesion and more convenient positioning of cerebral protection [4]. In case of PPHA originating below the mandibular angle or an atherosclerotic plaque extending to the level of the PPHA origin, it is possible to perform ECEA with a Y-shaped incision. The vertebral artery was aplastic/hyperplastic or frequently ended in the posterior inferior cerebellar artery The posterior communicating artery at the side of the lesion was absent in 86% (13/15, with no data available for 9 cases). This confirms high risk of cerebral ischaemia during surgery and necessity to use an intraluminal shunt in cross-clamping of arteries. For prevention of critical ischaemia in the anterior and posterior portions of the brain during ECEA, different variants of intraluminal bypass grafting have been described (Fig. 7) [12, 19].

We decided to modify the T-shaped shunt in order to simultaneously perfuse, if necessary, the carotid and basilar basins. Cerebral oxygenation was monitored by means of cerebral oximetry. On cross-clamping, the values of saturation in the carotid basin decreased insignificantly, therefore we used the shun only in the PPHA.

The literature has described different variants of distal embolic brain protection and their combinations in CAS (Fig. 8) [4].

CONCLUSIONS

The presence of persistent primitive arteries is associated with an increased risk for the development of cerebrovascular aneurysms, combined neurovascular syndromes, atherosclerotic lesions and ACI in the carotid and basilar basins. It is necessary to thoroughly identify a persistent anastomosis prior to planning of therapeutic decision-making. Damage or thrombosis of the PPHA which is often the only source of blood supply of the basilar artery may during surgery result in catastrophic cerebral ischaemia. PPHA with associated atherosclerotic disease is a difficult and potentially dangerous condition given the concomitant underdevelopment of carotid-basilar anastomoses and vertebral arteries. An approach to treatment of this pathology should be individual. Surgical treatment, although feasible and successful in reported cases, can pose significant risk. Limited reports show a possibility of successful endovascular treatment.

Conflict of interest: none declared.

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