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RELEVANCE OF PRIMITIVE CAROTIDOBASILAR ANASTOMOSIS FOR INTERNAL CAROTID ARTERY STENOSIS

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Primitive carotido-basilar anastomoses (PCA) are persistent fetal vessels.
The aim of the study was to compare the clinical characteristics of patients operated on for internal carotid artery (ICA) stenosis with or without PCA in order to evaluate the impact of PCA on the treatment.
Material and methods. Consecutive patients operated on for ICA stenosis at our university hospital were included. Surgical treatment consisted in carotid endarterectomy (CEA) with patch plastic.
Results. Of the 380 CEA performed between 2006 and 2012, PCA were found in six patients (1.6%). All patients with PCA were symptomatic vs. 54% of patients without PCA (p=0.035). Significantly less posterior collateral flow was present in patients with PCA (33%) compared to those without PCA (85%, p=0.01). Only two of the six patients with PCA were diagnosed prior to surgery, none was ligated intraoperatively. PCA was not associated with stroke and restenosis at long-term follow up.
Conclusions. PCA are rarely diagnosed prior to surgery in patients with ICA stenosis and need not to be ligated during CEA.
Key words: internal crotid artery stenosis, primitive carotidobasilar anastomosis

Persistent primitive carotido-basilar arteries (PCA) are remnants of the early embryological anastomoses between the longitudinal arteries along the basal surface of the hindbrain, namely of the trigeminal, otic, hypoglossal and proatlantal intersegmentary arteries (1). They accompany the respective brain nerves and usually involute between the 30th and 40th day of fetal live when the posterior communicating artery forms (2). The persistent primitive trigeminal artery has a radiological incidence of 0.1-0.6% at the Magnetic Resonance Angiography (3), other variants are even less frequent. An association with other congenital conditions is often reported, such as moya moya disease (4), atrial septum defect (5), PHACE syndrom (6), hypo- or aplasia of the vertebral arteries or the proximal basilar artery (3) or lacking of the posterior communating cerebral artery (2). Various complications may result from these aberrant vessels, such as ruptured aneurysms with subdural hemorrhage (7), trigeminal neuralgia (8) or thromboembolism (9). On the other hand, in case of a carotid bifurcation stenosis, the PCA may serve as collaterals draining into the distal internal carotid artery, especially if associated with a hypoplastic vessel of the Circulus Willisii (2). Although there exist about 130 reviews, clinical investigations and case reports related to PCA in the Medline database to date, little is known about the contribution of PCA to the pathogenesis of internal carotidobasilar sclerosis. Vice versa internal carotid artery (ICA) stenosis associated with PCA may lead to ischemia of both the anterior and posterior cerebral territories, when the vertebral artery or the basilar artery are hypo- or aplastic (10).

Thus one of the aims of our investigation was to assess whether the clinical presentation of those patients carrying these anatomical variants differed from non-carriers. We analysed the clinical characteristics of all patients

operated on for ICA stenosis within a defined interval in our hospital and registered at the federal carotid surgery quality assurance database.

MATERIAL AND METHODS

Patients

From January 2006 until August 2012 all patients who were operated on at the Department of Vascular Surgery of our university hospital, for ICA stenosis were included. Patients who revealed a PCA were included into further analysis after a written consent was obtained. The preoperative anamnesis and physical examination as well as the routine laboratory investigation and the pre- and postoperative duplex-sonography of the carotid arteries were assessed. Furthermore, the intra-operative findings were documented by a written protocol, photography and digital subtraction angiography (DSA). The patients were followed-up after postoperative discharge in an ambulatory setting by a 6 week, half year and then yearly clinical and ultrasonographic examination.

Surgical procedure

The indication for surgery was determined by the symptoms, the severity of the ICA stenosis and its progress according to the national guidelines. Patients with elective or urgent operative indication underwent surgery with informed consent (corresponding to the legal requirements of the Federal Republic of Germany) in regional anesthesia including continuous monitoring of focal neurological status and vigilance. The incision was made along the M. sternocleidomastoideus on the respective side. The common carotid artery (CCA), the carotid bifurcation, the ICA and the external carotid artery (ECA) including the first level branches were identified and prepared. The patient was then given 5000 international units of unfractionated heparin intravenously. Before and after clamping of the CCA, the patient was asked to continuously press and release a squeaking duck with the opposite hand. When acute hemiparesis or a vigilance deficit developed, a temporary carotid shunt was inserted. The endarterectomy was performed after additional clamping of the distal branches of the bifurcation and longitudinal incision of the CCA. The distal intima was then fixed by 3-4 stitches with a 6-0 prolene suture, and the longitudinal incision was closed using a PTFE or XenoSure (LeMaitre) patch with a 5-0 Prolene running suture. The ICA clamp was released first, then the ECA clamp and then the CCA clamp whereby transiently pressing the ICA in order to conduct any remaining thrombus into the ECA. The wound was closed after introduction of drainage using an inversed subcutaneous single stitch suture with Vicryl 3-0 and intracutaneous resorbable running suture. The patient was monitored for at least one night at the Stroke Unit of the Neurological department depending on the general and neurological state. The Easy-flow drainage was removed on the next day and the postoperative ultrasonography of the cervical vessels was in general done the 4^{th}-5^{th} postoperative day after drainage removal. The patient was usually discharged on the 5^{th}-6^{th} postoperative day.

Statistics

Statistical calculations were done using the R statistical software (11). Scalar variables underwent comparison with t-test (if normally distributed) or Mann-Whitney-U-Test (if not normally distributed). Cross-tables were tested with Fisher´s exact test. The results were considered significant if $p<0.05$.

Ethics

Patients were only included if they gave their written informed consent (in case of death, the consent of the family was obtained). Patients with PCA were in addition asked for allowance to use anonymous image material and data for graphical and tabular presentation. The data security measures corresponded to the local legal requirements. Following the guidelines of the local Ethics Committee no further statement or decision concerning the performance or publication of the investigation was necessary due to its observational character.

RESULTS

From January 2006 until August 2012, 380 patients were operated on for ICA stenosis. Six of them revealed a carotido-basilar anastomosis (1.6% of all patients). The clinical characteristics of these patients are listed in tab. 1.

All patients were symptomatic, but were successfully treated and did not suffer from a persisting postoperative neurologic deficit. In only 2 instances could the PCA be diagnosed preoperatively by duplex sonography when coinciding with hypoplasia of the vertebral arteries. In three cases the PCA was observed only during surgery, because the aberrant arteries were small and not looked for by sonography (preoperative magnetic resonance angiography was not routinely performed). In case 1 (tab. 1) the PCA was diagnosed only at surgical revision: the patient showed a transient aphasia immediately after surgery and underwent computer tomography were the diagnosis of ICA dissection was suspected. Therefore the patient was then directly transferred to the operation theatre and revised. Nevertheless, no dissection was found, but a proatlantic/hypoglossic artery, which had remained obviously patent after the first operation and was perfused in an antegrad fashion. Case 2 presented with occlusion of the proximal right ICA and distal refilling via a PCA as assessed by duplex sonography. The preoperative DSA confirmed this finding and revealed additional distal occlusion of the right vertebral artery. After surgery the computer-tomographic angiography revealed a patent PCA with sonographically antegrad perfusion (fig. 1). The patient deceased after 30 months of ambulatory follow-up from unknown reasons. Nevertheless no re-stenosis of the right ICA was observed during follow-up.

When comparing the 1.6% incidence of PCA in our patients undergoing ICA surgery with those undergoing MRA for different reasons [0.1-0.6% (3), 0.1-0.5% (1), 0.33% (12)], the difference was significant (binomial test, p=0.0248, 95% confidence interval: 0.58 – 3.4%). However, with regards to the low percentage of preoperative diagnosis of PCA, MRA does probably not represent the true incidence of PCA. Therefore it would not be possible to draw any conclusions concerning the pathophysiology of ICA stenosis based on this difference.

We assessed furthermore whether the clinical courses revealed any differences in patients with and without PCA. The characteristics are listed in tab. 2. It is obvious that all patients with PCA were symptomatic while only 54% of those patients without PCA revealed ischemic symptoms (-test, p=0.035). All other parameters did not significantly differ.

In order to assess whether the difference in the percentage of asymptomatic ICA stenosis may be explained by altered collateralization of the extra- and intracranial blood supply, the vertebrobasilar system and anterior and posterior collateralization were quantified (tab. 3). The axial diameter of ipsi- and contralateral vertebral arteries (VA) were measured in the intervertebral (V2) segments, and hypoplasia was postulated if this diameter was less than 25 mm. One patient with PCA was identified with ipsilateral VA hypoplasia (17%).

Table 1. Clinical characteristics of patients with PCA.*Coincidence with a hypoplasia of the ipsilateral A. vertebralis

No	Gender	Age	Side	Grade %	Symptoms	Shunt	PCA	Diagnosed by	Hospitalization	Postop course	Follow-up	Current state
1	m	79	left	90	hemiparesis, aphasia	yes	n.n.	intraoperative DSA	14	revision	?	?
2	m	69	right	100	amaurosis fugax	no	A. proatlant.*	duplex sonography	8	uneventful	30	deceased
3	f	60	right	90	amaurosis fugax	no	n.n.	intraoperative	6	uneventful	6	well
4	m	75	left	90	TIA, aphasia	no	n.n.	intraoperative	12	uneventful	2	well
5	m	60	right	90	hemiparesis	yes	n.n.	intraoperative	7	uneventful	0	well
6	m	37	left	100	hemiparesis, aphasia	?	A. proatlant.*	duplex sonography	?	hemiparesis	2	no deficit

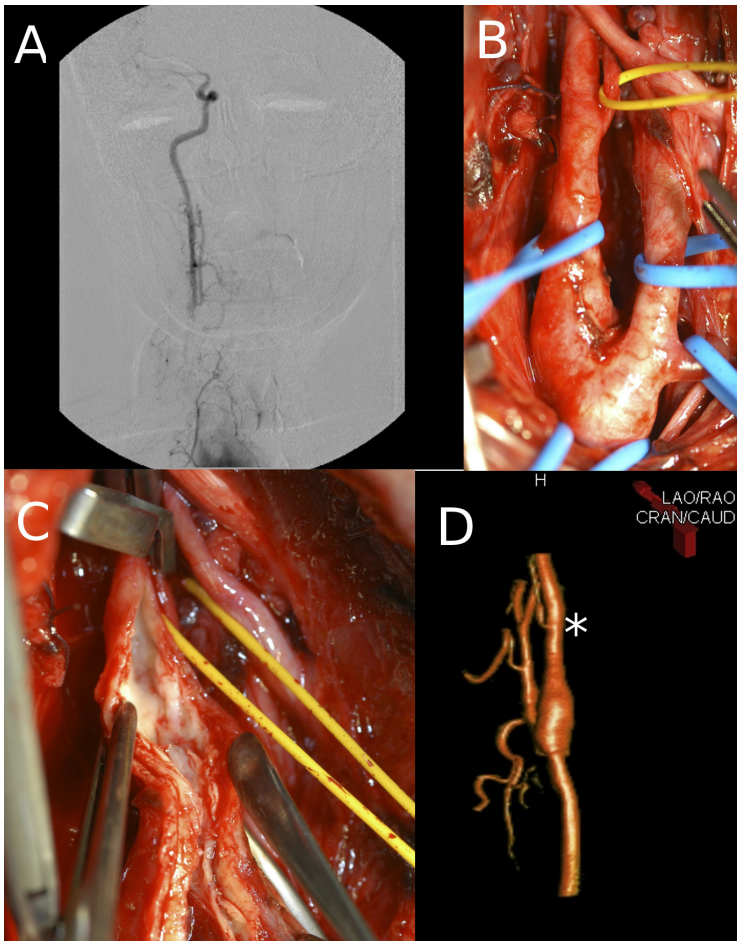

Fig. 1. Radiologic and intraoperative demonstration of the PCA in case 2 (see tab. 1)

A: preoperative DSA showing occlusion of the right ICA and refilling via a distal branch, which was identified as PCA and retrogradly perfused. B: intraoperative finding – CCA, ICA ECA and superior thyroid artery were marked by blue vessel loops. The carotid bifurcation can be easily identified and the PCA is branching off distally (yellow vessel loop). C: incised after clamping bifurcation (not shown) and proximal ICA, which is completely occluded by an arteriosclerotic plaque. The more distal ICA is stenotic but perfused from the level of the branching off of the PCA. D: postoperative 3-dimensional CT-angiography reconstruction. The persistent, but now supposedly antegradly perfused PCA is marked at the level of branching off the ICA by a star (latero-dorsal perspective)

Nevertheless, the difference to the cohort without PCA (7%) was not significant (p=0.36). Moreover, when both VA were summarised, and a sum of axial diameters less than 5 mm was defined as vertebrobasilar hypoplasia, none of the patients with PCA and only 1.4% of the patients without PCA carried this anomaly (p=1). Anterior collateral flow was postulated when the ipsilateral A1 segment of the anterior cerebral artery was perfused in retrograde direction. This was the case in 26% of patients without PCA but in none of those patients carrying the PCA (p=0.35). Posterior collateral flow was defined as perfusion of the ipsilateral posterior communicating artery in dorsoventral direction and was found in 33% of patients with PCA and in 85% of patients without PCA (p=0.01).

DISCUSSION

One of the aims of our investigation was to assess whether PCA increased the risk for ICA

Table 2. Characteristics of patients with and without PCA. The percentage of asymptomatic patients in the group w/o PCA is significantly higher than the percentage in those patients w/PCA

Variable	With PCA	Without PCA	p
Mean age (years)	67	69	0,89*
Asymptomatic (% of all)	0	46	0,035#
Recurrent stenosis (% of all)	0	1	1#
Ipsilateral stenosis (% of diameter)	80	84	0,35*
Contralateral stenosis (% of diameter)	26	35	0,56*
Mean interval (days)	34	22	0,10$

Abbreviations: * – Student´s t-test, # – ^{2} -test, $ – Mann-Whitney'a-U-test. Mean interval means time (in days) between the ischemic event and surgery in symptomatic patients

stenosis. A possible role for the hemodynamics at the level of the PCA´s branching off the ICA has been postulated in the past (13). Our investigation revealed that the incidence of PCA in patients undergoing ICA surgery is slightly and significantly higher than in those who underwent MRA from various reasons (3) (1.6% vs 0.6%, Binomial test). Nevertheless when comparing the diagnosis of PCA before surgery (only in two cases = 0.5%) with the MRA study (3), both incidences were nearly identical. Therefore, these variants may simply be underdiagnosed by routine preoperative diagnostics. Moreover, age at the time of diagnosis was not significantly different in patients with or without PCA, which would have pointed to an etiological contribution of PCA to ICA stenosis. We conclude that based on our data there is no evidence for a higher risk to develop ICA stenosis in patients carrying a PCA compared to normal population.

Furthermore both cohorts (with and without PCA) were compared considering the clinical presentation, because PCA are often associated with other cerebrovascular anomalies, such as aplasia of the posterior communicating artery or hypoplasia of one or both of the vertebral arteries, reducing the compensatory capacities of the Circulus Willisii. All patients carrying a PCA were symptomatic while patients without PCA were asymptomatic in 46% (tab. 2). This difference was significant (p=0.035, Fisher´s exact test). Their symptoms, however, did not differ from those of the majority of patients without PCA (tab. 1). Therefore, further investigations aimed at comparison of collateralization.

As shown in tab. 3, hypoplasia of the ipsilateral VA occurred slightly more often in patients with PCA (n=1, 16.7%) than in patients without PCA (7%). However this difference was not significant (p=0.36, Fisher´s exact test). In addition, when comparing the sum of the axial diameters of both VAs, insufficiency of the whole vertebrobasilar system was exceptionally rare (0% in patients with PCA, 1.4% in the "control" cohort). In most cases of ipsilateral hypoplasia, it is compensated by a contralateral hyperplasia. The retrograd flow via anterior communicating artery representing the contralateral collateralization was not present in patients with PCA, but in 26% of those patients without PCA (p=0.35). Furthermore the vertebrobasilar collateralization via posterior communicating artery (dorsoventral flow) could be observed in 33% of patients with PCA, but in 85% of patients without PCA (p=0.01). This might indicate that the collateralization is worse in patients with PCA compared to those without PCA, possibly due to additional variants or anomalies, such as aplasia of the anterior communicating artery. Microembolizations from emboligenic ICA plaque may also play a role in pathogenesis of neurologic symptoms, but detection by intracranial Doppler ultrasounds influenced by the current pharmaceutical thrombocyte aggregation inhibition which was initiated in almost all patients with symptomatic ICA stenosis at the time of admission. Moreover it would be difficult to draw a causal line from PCA to emboligenic plaques. Therefore we did not compare microembolization rates of patients with and without PCA.

Table 3. Comparison of patients with and without PCA using characteristics of collateralization

Measurement	PCA (n=6)	Control cohorts	p*
VA ipsilat. hypoplastic	1 (16,7%)	15/218 (7%)	0,36
VB hypoplastic	0	3/210 (1,4%)	1
Anterior collateral flow	0	54/210 (26%)	0,35
Posterior collateral flow	2 (33,3%)	45/53 (85%)	0,01

* Fisher´s exact test

Surgery in regional anesthesia may be favorable because neurological deficits after ICA clamping can be more easily recognized in case of insufficient collateralization. Probably in most cases, the PCA cannot be diagnosed before surgery, so it remains the surgeon´s responsibility to consider such a variant and to identify the ICA correctly (because of the fact that usually it is identified as the vessel who does not branch). The PCA may not be ligated, although after surgery, it may spontaneously obliterate without serious sequelae. However this does not concern those cases when patients present with primary complications of these aberrant vessels, such as aneurysms, trigeminal pain or embolism. An intraoperative temporary carotid shunt is

usually not necessary as long as the patient does not deteriorate.

Based on our findings, it seems reasonable to leave the PCA untouched, when found before or during operation, and to perform further investigation (CTA or MRA) in order to exclude coincidental cerebrovascular anomalies.

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