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Postpartum Cervicocephalic Artery Dissection

Marcel Arnold, MD; Mathilde Camus-Jacqmin, MD; Christian Stapf, MD; Anne Ducros, MD, PhD; Anand Viswanathan, MD, PhD; Karine Berthet, MD; Marie Germaine Bousser, MD

Background and Purpose—Cervicocephalic artery dissection (CAD) after childbirth is rare. The objective of this study was to determine differences between postpartum and nonpostpartum CAD.

Methods—We compared consecutive patients with postpartum CAD with a control group of women with nonpostpartum CAD.

Results—Of 245 patients with CAD, 102 women <50 years (6 with postpartum CAD and 96 with nonpostpartum CAD) were identified. Vascular risk factors and presenting characteristics did not differ significantly between postpartum CAD and nonpostpartum CAD women. By contrast, patients with postpartum CAD had more often coexisting conditions such as reversible cerebral vasoconstriction syndrome (2 of 6 versus 2 of 96; $P=0.017$), reversible posterior leukoencephalopathy syndrome (2 of 6 versus one of 96; $P=0.009$), and subarachnoid hemorrhage without signs of intracranial extension of CAD (2 of 6 versus zero of 96; $P=0.003$).

Conclusion—CAD and associated conditions should be looked for in women with unusual headache after childbirth. (*Stroke*. 2008;39:2377-2379.)

Key Words: stroke ■ childbirth ■ dissection ■ postpartum angiopathy
■ reversible cerebral vasoconstriction syndrome

Postpartum cervicocephalic artery dissection (ppCAD) is a rare and still poorly understood condition. We add to the 15 cases so far reported¹ 6 cases prospectively recruited in the last 10 years.

Methods

Patients were recruited from an ongoing monocenter prospective cervicocephalic artery dissection (CAD) registry from January 1997 through October 2005. Inclusion criteria were as follows: female sex; aged <50 years; and diagnosis of spontaneous CAD by catheter angiography or MRI. Risk factors were assessed as reported before.² ppCAD was defined as CAD occurring within 6 weeks after childbirth.³ Clinical and radiological data were compared between ppCAD and nonpostpartum CAD women. Outcome at 3 months was assessed using the modified Rankin scale.

Statistical Analysis

For differences between postpartum and nonpostpartum women, χ^2 test, Fisher exact test, or Mann-Whitney test were used as appropriate. For comparison of outcome, we divided patients into 2 groups: modified Rankin scale 0 to 1 and modified Rankin scale 2 to 6.

Results

Among 245 patients with CAD, 102 women <50 years of age were identified among whom 6 had ppCAD (6%). Four had unilateral vertebral artery dissection (VAD) associated in one with internal carotid artery dissection (ICAD). One had bilateral VAD and one unilateral ICAD.

Baseline characteristics are summarized in Table 1. Headache and/or neck pain were the initial symptoms in all

patients and developed between 5 and 18 days after delivery. Headache was bilateral in 4 patients and unilateral in one. It qualified for thunderclap headache in 2. Headache was the only sign of CAD in 2 patients. It was associated with Horner syndrome in one, with transient ischemic attacks in 2, and ischemic stroke in one. Five women had a history of migraine and hypercholesterolemia.

All migrainous women reported that their headache was different from their previous migraine attacks. None had a history of hypertension but 3 had a transient increase in blood pressure postpartum. The outcome was favorable (modified Rankin scale 0.1) in all 6 women.

The comparison of ppCAD with nonpostpartum CAD is indicated in Table 2. The only significant difference is that women with ppCAD had more often coexisting conditions such as reversible cerebral vasoconstriction syndrome (RCVS; 2 of 6 versus 2 of 96; $P=0.017$), reversible posterior leukoencephalopathy syndrome (2 of 6 versus one of 96; $P=0.009$), and subarachnoid hemorrhage (SAH) without signs of intracranial extension of CAD (2 of 96 versus zero of 96; $P=0.003$).

Discussion

Although women are at a higher risk of stroke postpartum, ppCAD is rare with altogether 15 cases reported so far, 9 with unilateral ICAD, one with bilateral ICAD, one with triple CAD, and one with dissection of all 4 cervical arteries.^{1,3-7} In addition, 2 patients with basilar artery dissection have been described.⁸

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Table 1. Characteristics of Women With ppCAD

Patient	Age, Years	Dissected Vessels	Potential Risk Factors and Triggers	Symptom Onset, Days After Delivery	First Symptom	Other Clinical Manifestations	Associated Conditions
1	41	VA left	Hypercholesterolemia Migraine	18	Bilateral neck pain	TIA	
2	35	ICA left VA right	Hypercholesterolemia Migraine, smoking, elevated blood pressure Bromocriptine Zolmitriptan	5	Bilateral headache	Ischemic stroke	RCVS
3	38	ICA right	Hypercholesterolemia Migraine, elevated blood pressure Urinary tract infection	8	Bilateral headache	Horner syndrome	SAH
4	27	VA right	Hypercholesterolemia Migraine	11	Ipsilateral neck pain, TCH		RCVS, RPLS
5	38	VA right VA left	Hypercholesterolemia Migraine, elevated blood pressure, fever	7	TCH	TIA	SAH, RPLS
6	34	VA right	Chiropractor neck manipulation	7	Ipsilateral neck pain headache		

VA indicates vertebral artery; ICA, internal carotid artery; TCH, thunderclap headache; TIA, transient ischemic attack.

We report 6 women with 8 ppCAD (6 VAD and 2 ICAD). The mechanism of these ppCADs and their relationship with delivery remain unclear; except for one patient who had chiropractic manipulation, traumatic dissection triggered by a strenuous effort during labor seems unlikely in these women who had uneventful vaginal deliveries and in whom there was an interval of over 5 days between delivery and symptom onset. Furthermore, all women also had uneventful pregnancies without signs of eclampsia. It is however possible that the transient increase in blood pressure observed in 3 women postpartum had played a role.

Among underlying conditions, 5 women had a history of migraine and hypercholesterolemia. Although migraine has been found to be an independent risk factor for CAD in 3 case-control studies,⁹ there is no evidence so far that hypercholesterolemia is a risk factor for CAD.

We observed no significant difference between ppCAD and nonpostpartum CAD regarding clinical presentation and outcome. Headache and/or neck pain was the initial symptom in all patients. It was followed by ischemic events in 3 (transient ischemic attacks in 2, cerebral infarction in one), associated with a Horner syndrome in one, and completely isolated in 2. Although rare, CAD should thus be added to the long list of causes of headache postpartum, even in the absence of Horner syndrome or of cerebral ischemic events.

The most striking finding in our series is the presence in 4 of 6 patients with ppCAD of associated conditions such as RCVS, reversible posterior leukoencephalopathy syndrome (RPLS; Figure), and SAH (Figure), which, by contrast, were rare in ppCAD affecting only 3 of 96 patients. RCVS occurring postpartum, reported as postpartum angiography, is characterized by pro-

longed but reversible vasoconstriction of cerebral arteries, usually associated with acute-onset, severe, recurrent headaches with or without associated manifestations such as seizures or ischemic or hemorrhagic stroke.^{10,11} RCVS may also coexist with RPLS both during and outside postpartum¹² and may be associated, in as many as 20% of cases, with a small localized cortical SAH.^{11,12} We have recently shown that, first, when present in RCVS, both RPLS and cortical SAH were early events occurring in the first few days, and second, that the typical arterial narrowing may not be visible on an initial angiogram.¹¹ In our present series, among the 4 patients with associated conditions, 2 had an angiographically documented RCVS, isolated in one and associated in the other with RPLS. Another patient had RPLS associated with a small localized cortical SAH and the fourth patient had a localized cortical SAH. Both had normal angiograms. Nevertheless, an underlying RCVS is also the most likely explanation in these 2 patients despite the normal angiogram. Thus, among our 6 patients with ppCAD, 2 had definite postpartum angiography and 2 had probable postpartum angiography. Associated risk factors for RCVS were present in only one patient who received bromocriptine to stop lactation and zolmitriptan, a vasoconstrictive anti-migraine drug.

To date, the association of CAD and postpartum angiography has been reported in only one case and was considered coincidental.¹² Our series suggests that there is a relationship between these 2 conditions, but the mechanisms underlying this association remain unknown. Furthermore, in our patients as well as in the only other reported one, it is impossible to know which of the 2 conditions occurs first, because both present with acute severe headache. One might speculate

Table 2. Vascular Risk Factors, Presenting Characteristics, and Concomitant Conditions

Characteristic	Postpartum Dissection (n=6)	Nonpostpartum Dissection (n=96)	P Value
Mean age, (SD), years	35.4 (4.5)	38.7 (7.3)	0.28
Dissected vessels			
Carotid artery	1	57	
Vertebral artery	4	33	0.11
Both	1	6	
Multiple dissections	2	23	0.63
Hypercholesterolemia	4	41	0.15
Hypertension	0	6	1.0
Mean systolic blood pressure on admission (SD)	135 (39)	130 (22)	0.87
Mean diastolic blood pressure on admission (SD)	78 (17)	77 (14)	0.82
Diabetes	0	1	1.00
Current smoking	1	26	1.00
Oral contraceptives	0	30	0.17
Hereditary connective tissue disorder	0	2	1.00
Family history of stroke	0	15	0.59
Migraine	5	47	0.21
Minor trauma (other than delivery)	1	14	1.00
Presenting symptom			
Stroke	1	46	
Transient ischemic attack	2	17	
Retinal ischemia	0	4	0.40
Local symptoms only	3	29	
Headache	6	80	0.59
Neck pain	5	51	0.22
Tinnitus	0	16	0.59
Cranial nerve palsies	0	2	0.80
Horner syndrome	1	27	0.84
Time to diagnosis			
Mean (SD), days	11 (8)	8 (7)	0.16
RCVS	2	2	0.017
RPLS	2	1	0.009
SAH	2	0	0.003

about the role of transient arterial wall abnormalities and/or increases in blood pressure induced by postpartum hormonal changes. Migraine might also play a role because all the patients with ppCAD and nonpostpartum CAD with both conditions in our series had migraines.

In conclusion, ppCAD accounts for 6% of spontaneous CAD in women under 50 years of age. There is a significant association of unknown mechanism between ppCAD and postpartum angiography (RCVS). Both conditions should be looked for when women experience unusual severe headaches after childbirth because they require different treatments.

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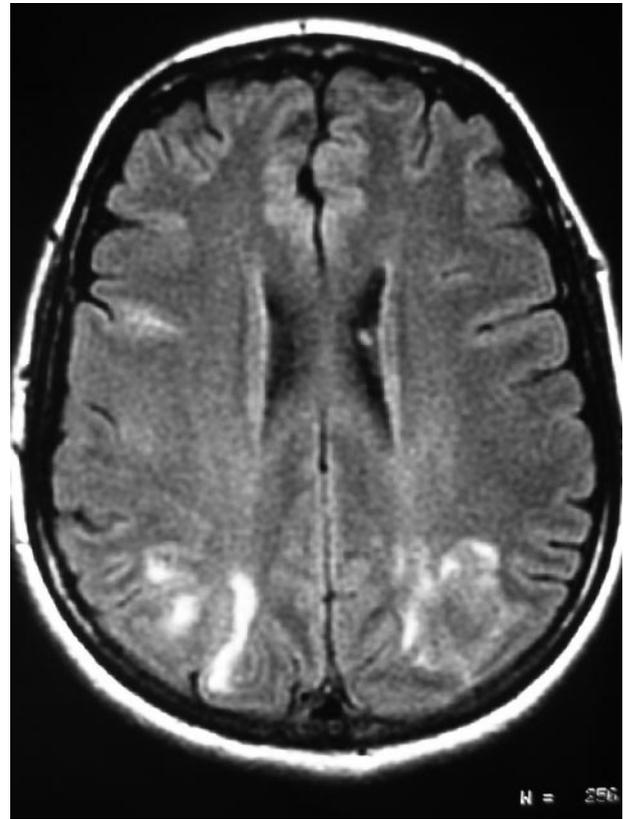


Figure. MR brain imaging of Case 5 showing posterior white matter lesions compatible with postpartum RPLS and SAH into the intersulcal space over the left hemisphere.

Disclosures

None.

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