Death in a postpartum woman caused by an exceptionally rare case of concurrently rupture of bilateral non-traumatic carotid-ophthalmic aneurysm associated with bilateral acute subdural hematoma

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Abstract: Nontraumatic intracranial aneurysms have a 2-5% incidence in the population and most are asymptomatic. However, even though the risk of rupture is low, it has a high mortality and morbidity. We report an extremely rare case of concurrently rupture of bilateral nontraumatic carotid-ophthalmic aneurysm complicated with acute bilateral subdural hematoma in a postpartum woman. The imagistic examinations showed two saccular bilateral aneurysm located at the carotid-ophthalmic junction, bilateral basal frontotemporal subdural hematoma, a right frontotemporal intraparenchimatous hematoma and massive cerebral edema. Additionally, the autopsy revealed the location of rupture of both aneurysms, and the presence of recent bilateral thrombi in the lumen of the cervical segment of the internal carotid arteries (as a result of the mechanical occlusion of these arteries due to the massive cerebral edema). In the absence of any medical and antenatal check-ups records of the patient, we concluded that in the pathogenesis of the development and rupture of these aneurysms could contributed the following factors: hemodynamic, blood, and arterial wall changes during pregnancy and puerperium, infection, presence of intraluminal aneurysm thrombus. We have suggested the possible pathogenic mechanism for the bilateral subdural hematoma based on the hypothesis of the aneurysm sentinel bleeding, sustained by the patient's symptomatology.

Key Words: bilateral ruptured carotid-ophthalmic aneurysm, bilateral acute nontraumatic subdural hematoma, puerperium, sentinel hemorrhage.

Non-traumatic intracranial aneurysms represent an acquired pathology found in 2 - 5% of the population [1]. Most are asymptomatic especially if they are small in size but all intracranial aneurysms (despite of their size) have the potential for rupture, frequently causing subarachnoid hemorrhage and rarely intraparenchimatous brain hematoma, with a high mortality (50 – 60% of the cases [1]) and morbidity rates (20 – 25% of the survivors are affected by permanent neurological complications [1]).

The eye globe and adnexa are irrigated mainly by the ophthalmic artery (OA) that has a major importance for vision through its central retinal artery that nourishes the retina. In most cases OA branches from the superior-medial wall of the internal carotid artery (ICA) [2], as it emerges from the roof of cavernous sinus medial to

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anterior clinoid process of the sphenoid bone, close to the optical canal [3], having a diameter of approximately 1.5mm [4-6].

According to Bouthiliers classification [7] this region where the OA has its origin represents the 6th segment of the ICA, known as the ophthalmic segment that extends from the distal dural ring to the posterior communicating artery [7-9], having an average length of 9.6 mm [10]. The superior hypophyseal artery also emerges from the same segment [9]. From anatomical point of view, the OA has the following segments: intracranial, that has 0.5 – 9.5mm in length [11] where the artery is located below the optic nerve and presents one or two angles [11]; intracanalicular, having a length of between 5 – 7mm [11] where the artery runs inferolateral to the optic nerve [2, 3, 11, 12]; intraorbital, in the first part of this segment the artery continues the inferolateral course to the optic nerve [2, 11, 12], then in the second part the artery crosses above (in most of the cases) or below the optic nerve [2, 11, 12] and in the third part it runs tortuously along the medial wall of the orbit [3]. The OA gives off the following branches: central retinal, lacrimal, ciliary, supraorbital, posterior and anterior ethmoidal, dorsonasal, supratrochlear and medial palpebral arteries [3, 12].

Aneurysms that originate in the ophthalmic segment at or around the emergence of the ophthalmic artery are known as carotid-opthalmic aneurysms [13-16] or more commonly, just ophthalmic aneurysms [9, 13, 17]. The later nomenclature could create misunderstandings, these morphoclinical entities should not be confused with the aneurysms originating in the ophthalmic artery trunk and its branches which are known as peripheral ophthalmic artery aneurysms [13], or “true” OA aneurysms [15].

We report and discuss the pathogenesis of an extremely rare case of a concurrently ruptured non-traumatic bilateral carotid-opthalmic artery aneurysms associated with acute bilateral subdural hematoma in a postpartum woman.

CASE REPORT

A 31 years old woman in her second week after birth (vaginal delivery) was found unconscious and according to her family she had complained earlier in the day of an atrocious headache associated with explosive vomiting, after a few days of progressive headaches with no visual disturbances. The family stated that the patient had no known pathological history and also no past medical or antenatal check-ups records were available. On admission in the ER, the patient had a GCS of 3 with anisocoric pupils (right pupil larger than the left), and a blood pressure of 180/100mmHg. The CT brain scan showed the following: a right frontotemporal intraparenchimatus hematoma (measuring 59/53mm transaxial and 51mm craniocaudal) compressing the right lateral ventricle; a bilateral basal frontotemporal subdural hematoma with a maximum thickness of 15mm frontal left and 4mm frontal right; diffuse cerebral edema with effacement of the basal cisterns, and a tendency for cerebellar tonsils herniation and a 9mm midline shift to the left. Due to the imminent, life-threatening nature of the patient's pathology, a right frontal decompressive craniectomy was immediately performed and the frontotemporal intracerebral hematoma (measuring approximately 75/5/4cm) and also a 5mm thick right frontotemporal subdural hematoma were evacuated. After the neurosurgical intervention, the patient presented a GCS of 3, no brainstem reflexes, fixed mydriasis, with hemodynamic instability on Noradrenaline support and she was mechanically ventilated. Next morning, in order to find the source of bleeding a bilateral carotid angiography was performed, which showed two saccular aneurysms located bilaterally at the carotid-opthalmic junction, the right aneurysm (Fig. 1) measuring 13/8mm (a large aneurysm according to ISUIA classification [18]), the left one (Fig. 2) measuring 8/6mm (a small aneurysm according to ISUIA classification [18]), both superiorly oriented, presenting a smooth contour and a neck of approximately 3mm.

The patient’s condition remained critical with no recommendation for surgical intervention and despite the intensive care provided, the patient died on the fourth day after admission in the hospital. A medico-legal autopsy was requested to determine the exact cause of death and to eliminate any suspicion of a violent death.

Autopsy findings

The external examination of the body revealed a frontotemporoparietial arcuate sutured surgical incision and no traumatic injuries of the scalp and body. The internal examination showed the following relevant findings: no skull fractures, absence of the bone flap, diffuse and massive cerebral edema with flattening of gyri and obliteration of sulci, significant cerebellar tonsillar herniation, brainstem hemorrhage, diffuse subarachnoid hemorrhage (with a maximum thickness along both frontotemporal basal regions of the brain); subdural hematoma on the left middle temporal fossa and thin subdural hematoma on the left anterior fossa, the right ICA was cut during autopsy immediately after its emergence from the cavernous sinus under the origin of the ophthalmic artery and presented a sacciform dilatation (Fig. 3) of approximately 14/8mm with an inflammatory aspect, having an irregular surface, an intraluminal thrombus and a rupture of about 2mm (Fig. 4) located at the origin of the ophthalmic artery; the left ICA was cut approximately 1cm after its emergence from the cavernous sinus, the dilatation was smaller in size than the right one and presented an intra vitam rupture of approximately 2mm located at the ophthalmic artery.
origin, with irregular and hemorrhagic infiltrated margins (Fig. 5, yellow arrow), characteristics that are not found in the autopsy artefact indicated in the figure 5 by the blue arrow; an adherent thrombus to the arterial wall was found almost completely occluding the lumen of each ICA (Figs 6 and 7), extending throughout the entire cervical segment, starting from the bifurcation of the carotid artery (we could not determined if the thrombi extended up in the other segments of the internal artery carotid).

The histopathological examination of the specimens collected at autopsy, revealed the following: brain - recent massive subarachnoid hemorrhage, accentuated cerebral edema; carotid-ophthalmic aneurysm – atherosclerotic mural changes, firmly adherent mural thrombi consisting of fibrin, erythrocytes and leukocytes, moderate perivascular hematic infiltrate; ICA – the lumen is almost completely occluded by a recent thrombus consisting of fibrin, erythrocytes and leucocytes; lungs – bronhopneumonia; uterus - acute suppurative endomyometritis.

Figure 1. Right carotid-ophthalmic aneurysm: (A) anteroposterior view, (B) lateral view.

Figure 2. Left carotid-ophthalmic aneurysm: (A) anteroposterior view, (B) lateral view.
DISCUSSION

The carotid-ophthalmic aneurysms [16] represent 1.3-5% of all intracranial aneurysms [10] and have a lower risk of rupture than other intracranial aneurysms [9, 16] (the annual average risk of rupture for intracranial aneurysms with a diameter between 7 – 12mm is 0.5% and 1% for aneurysms between 13 – 24mm [19]).

Considering these aspects, our reported case has extremely rare features consisting of a concurrently rupture of bilateral carotid-ophthalmic artery aneurysms complicated with unilateral intraparenchimatous hematoma and bilateral acute non-traumatic subdural hematoma associated with subarachnoid hemorrhage. In the consulted literature there is only one report of bilateral acute non-traumatic subdural hematoma caused by a ruptured posterior communicating artery aneurysm [20] and 5 cases of simultaneously ruptured aneurysms.
originating in other intracranial arteries [21, 22]. The incidence of unilateral acute subdural hematoma from aneurysm rupture is just 0.9% [23].

It is well known that the presence of intracranial aneurysms is higher in women. A relatively recent study [16] showed a higher incidence of the carotid-ophthalmic aneurysms in women (10:1 female-to-male ratio) than the incidence of other intracranial aneurysms (3:1 female-to-male ratio) [19, 24].

The majority of intracranial aneurysms are saccular, located almost exclusively at or near the relatively unsupported bifurcation sites of the cerebral arteries [2, 25-27] where hemodynamic stress (e.g., arterial hypertension) acts on a weak arterial wall caused by structural changes of the media and internal elastic lamina [2]. Additional factors that could contribute to the formation of the saccular intracerebral non-traumatic aneurysms are: atherosclerosis, familial predisposition, congenital defects of the arterial walls, excessive smoking and alcohol consumption, infectious emboli, inflammatory and immunological diseases and idiopathic [2, 19, 26].

An aneurysm ruptures when the wall stress produced by the intraluminal pressure exceeds the wall strength [28, 29]. In the reported case, the aneurysms rupture was probably caused by a contribution of factors, detailed below.

The aneurysm walls could be weakened by the arterial hypertension that occurs in the third trimester of pregnancy by a 50% increase of the blood volume with a proportional increase in the cardiac output [30] associated with arterial wall structural changes (hyperplasia of arterial wall smooth muscle and loss of the normal elastic fibre alignment [30]).

The risk of aneurysm rupture during labour is increased by the elevation of intracranial blood and cerebrospinal fluid pressure due to Valsalva manoeuvres required to conduct vaginal delivery [26, 31].

The risk of intracranial aneurysm rupture during labour is 2% and 8% [32] during the first 6 weeks postpartum [33]. Immediately after delivery the arterial blood pressure falls temporarily then starts to increase reaching a maximum 3 to 6 days postpartum [33]. The probable cause of this transient hypertension include: pain, the movement of tissue salt and water accumulated during gestation into the circulatory system, normalisation of vascular tone [33]. In other cases the postpartum hypertension represents the persistence of gestational or chronic hypertension [34].

In the case presented, we have no information regarding patient’s pathologic history or pregnancy evolution, however the high blood pressure (180/100mmHg) measured at the time of admission in the hospital indicates the presence of a hypertensive condition that could triggered the rupture of both aneurysms weakened by the factors mentioned above.

In our case, another risk factor for aneurysm wall fragility and eventually rupture was the presence in the right aneurysm of an intraluminal thrombus, demonstrated macroscopically and microscopically. Frosten et al. [28] explained the disruptive effect of the intraluminal thrombus on the aneurysm wall through two mechanisms: an active mechanism caused by thrombus released factors and a passive mechanism caused by limiting the blood irrigation of the wall cells inducing their death.

The uterine source of infection (acute suppurative endomyometritis) could had also contributed to the aneurysms weakening process.

The imagistic and autopsy findings showed that the right intraparenchimatos hemotoma was caused by the rupture of the right carotid-ophthalmic aneurysm.

The traumatic etiology of the bilateral subdural hematomas was excluded based on the absence of scalp and body traumatical lesions and skull fractures. Considering the presence of the progressive headache a few days before admission in the hospital that could be explained by a sentinel hemorrhage (leakage of blood from the aneurysm[19]), the most likely pathogenesis of the subdural hematomas is this sentinel hemorrhage that caused adhesion between the aneurysm and the arachnoid membrane and the final rupture occurred into the subdural space [23]. Another possible explanation of the blood accumulation in the subdural space is the rupture of the arachnoid membrane under a high pressured bleed [35, 36].

The pathogenesis of the bilateral ICA thrombi involves the mechanical occlusion of the internal carotid arteries by the high intracranial pressure due to the massive cerebral edema [37]. This in turn results in the stagnation of blood that would increase the thrombi formation in association with the postpartum characteristic hypercoagulability (in the early postpartum period there is an increased risk for thrombosis associated with blood hypercoagulability due to high levels of fibrinogen and other coagulation factors [38]).

The absence of visual disturbances in our reported case are explained by the clinically well tolerated even of large aneurysms [39]. Visual symptoms usually occur in optic nerve atrophy [39] by chronic compression of giant aneurysms [40].

References
Dogăroiu C. et al. Rupture of bilateral non-traumatic carotid-ophthalmic aneurysm associated with bilateral acute subdural hematoma


