Case report

Spontaneous subarachnoid haemorrhage from rupture of an anterior communicating artery aneurysm in a patient with pituitary macroadenoma

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ABSTRACT

The presence of a cerebral aneurysm in patients with pituitary adenoma is a rare event. Diagnostic suspicion may stem from magnetic resonance imaging, which should lead to complementary investigation. As for treatment, even in conditions in which there has been no previous bleeding, the simultaneous approach should be considered, prioritising the aneurysm most of the time.

The present report describes the case of a patient with a history of pituitary macroadenoma, who had undergone a partial transsphenoidal resection ten years earlier. Admission to our service occurred after a sudden headache followed by mental confusion. A cranial computed tomography showed subarachnoid haemorrhage and expansive suprasellar lesion. Cerebral angiography showed a saccular aneurysm of the anterior communicating complex. The patient underwent a surgical procedure for microsurgical clipping of the aneurysm and partial resection of the pituitary tumour. We have also included a brief review of the literature on this subject.

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Hemorragia subaracnoidea espontánea debida a ruptura de aneurisma en la arteria de comunicación anterior en un paciente con macroadenoma hipofisario

RESUMEN

La presencia de aneurisma cerebral en pacientes con adenoma pituitario es un evento raro. La sospecha diagnóstica se puede obtener con la resonancia magnética, que debería conducir a una investigación complementaria. En cuanto al tratamiento, incluso en condiciones en las que no ha habido ningún sangrado previo, el enfoque simultáneo debe ser considerado, priorizando el aneurisma mayor parte del tiempo.

El presente informe describe el caso de una paciente con antecedentes de macroadenoma hipofisario, que fue sometido a una resección transesfenoidal parcial diez años antes. La

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http://dx.doi.org/10.1016/j.neucir.2013.03.005
**Introduction**

The presence of a cerebral aneurysm in patients with pituitary adenoma is a rare event and its frequency has not yet been definitively established. However, the incidence of cerebral aneurysms in patients with pituitary tumors is believed to be greater than that of the general population. Pant et al. found an incidence rate of 5.4% for this association, demonstrating that 97% of these aneurysms were located in the anterior circulation and that in 12% were multiple. Wakai et al. state that in 7.4% of pituitary tumors there is an association with cerebral aneurysms. Also according to these authors, the association of cerebral aneurysms with pituitary tumors is more common than the coexistence with other cerebral tumors. These facts highlight the questioning as to the existence of a causal or favorable link between the same.

The articles that deal with the subject in their majority relate situations in which the coexistence occurs with unruptured aneurysms and leads to the modification of the diagnostic and therapeutic strategy. The present case study has the objective of presenting and reviewing the appropriate literature on an even more uncommon event: the occurrence of subarachnoid hemorrhage due to the rupture of the anterior circulation aneurysm in a patient who already has the diagnosis of pituitary macroadenoma.

**Case report**

A male, 53-year-old patient with a history of pituitary macroadenoma (prolactinoma) was submitted to a partial transsphenoidal resection at another service ten years prior to the current hospitalization. He had associated bilateral amaurosis and had missed neuroendocrinological follow-up for 6 years. He was admitted to the emergency unit at our service following a sudden very intense headache condition accompanied by mental confusion. Upon neurological examination, the patient presented confusion, Glasgow coma scale score 14, without appendicular motor deficits (Hunt and Hess grade III). His pupils were mydriatic and not photoreactive.

A cranial computed tomography (CT) was solicited and it showed subarachnoid hemorrhage (Fishier IV) in base cisterns and left sylvian fissure, as well as interhemispheric hemorrhage collection (Fig. 1A–C). In addition, the presence of suprasellar expansive lesion (Fig 1A–C) was observed. This formation was observed to be homogeneously contrast-capturing in the CT performed 7 days after the ictus (Fig. 1D–F). In view of these images and the history of pituitary tumor, two principal diagnostic hypotheses arose: tumor bleeding or hemorrhage due to the rupture of the partially thrombosed giant aneurysm. Therefore, a cerebral angiography was performed and it showed a saccular aneurysm in the anterior communicating complex, associated with a superior deviation in the anterior cerebral arteries (Fig. 2).

The patient was submitted to a surgical procedure in which right pteronal access had been opted for, followed by the microsurgical opening of the sylvian fissure and exposure of the anterior cerebral arteries and anterior communicating complex for the clipping of the aneurysm and partial resection of the pituitary tumor. The tumor was intimately adhered to the optic nerves and the chiasmus, as well as to the anterior cerebral arteries. The patient presented a good progression, without additional deficits, being classified in Glasgow outcome scale 4, due his previous amaurosis. He was once again referred to neuroendocrinological follow-up.

**Discussion**

The first register of an association between a cerebral aneurysm and a pituitary adenoma, documented by the French ophthalmologist Offret, dates back to 1959. However, to this day the joint incidence of these two disorders has not been completely determined, so it is possible to consider such association more frequent when comparing it with other primary tumors in the central nervous system. Pituitary adenomas, producers of the growth hormone, are the most commonly involved. Despite the fact that the prolactinoma is the most frequent pituitary tumor, the incidence of coexistent cerebral aneurysms seems to be lower in comparison with other adenomas, considering that approximately 2% of the prolactinomas are associated with cerebral aneurysms. However, in a recent retrospective study, which included 800 patients with pituitary adenoma, Min Chu Oh et al. observed no relationship between aneurysms and the tumoral histological type tumoral.

In the present case, we report on the association between a pituitary macroadenoma, producer of prolactina, and an anterior communicating artery aneurysm. The factors which determine this association are yet unknown, but there are situations in which the causal relationship is consistent. The epidemiological value on which this association is based rests on the fact that in the general population, according to the ISUIA study, aneurysms are located at a frequency of 89% in the anterior circulation and 11% in the posterior circulation. When associated with pituitary adenomas, 97% are found in the anterior circulation.

In 1990, Reddy et al. operated on a patient with a pituitary adenoma, producer of the growth hormone, who had a
normal cerebral angiography, by the transsphenoidal route. After five weeks, a new angiography showed a cavernous segment aneurysm in the internal carotid artery. Nagata et al. in 2010 reported a fusiform dilatation of the internal carotid artery two weeks after aggressive surgical resection of a suprasellar craniopharyngioma. This events illustrates the traumatic hypothesis as etiology for the formation of the aneurysm, a situation which cannot be excluded from the discussion of the present study, as the patient possessed a history of pituitary tumor transsphenoidal resection, despite not having previous angiographies.

Another factor involved in the formation of cerebral aneurysms in this context is that of mechanical alterations. The tumor growth leads to microanatomical alterations in the cerebral circulation which, in light of the compression or traction caused by the tumor, leads to a modification in the blood flow and, consequently, greater hemodynamic stress, thus favoring the formation of the aneurysm. According to Min Chu Oh et al., the most strongly factor related to the presence of a cerebral aneurysm in this context would be the cavernous sinus invasion, which precipitate such hemodynamic changes. In the same article, another variable with a statistical value that favor aneurysm formation is the age of the patient.

Hormonal alterations have also been mentioned as possible etiological factors, implying that such variations would lead to microcirculatory alterations, which in turn predispose the formation of the aneurysm. The growth hormone and associated growth factors play a fundamental role in the formation of the aneurysm and the fact that in some series approximately 50% of the pituitary adenomas associated with cerebral aneurysms are producers of the growth hormone supports these hypotheses. Another fact that supports this concept is found by Manara et al., that, in a prospective study including 153 patients with acromegaly, found that GH levels were related to the presence of cerebral aneurysms and suggested that one should consider conducting a neuroradiological investigation of the cerebral circulation when monitoring such cases.

The proximity of the tumor to the aneurysm makes the diagnosis more difficult and transforms the neurological treatment into a challenge, bearing in mind that ideally the two conditions should be treated. As was observed in this case, an expansive lesion having the effect of a mass associated with the subarachnoid hemorrhage may represent the expression of either the partially thrombosed giant aneurysm or a suprasellar tumor with bleeding. In the present study, with the presentation of an aneurysm after subarachnoid hemorrhage, the obvious priority was given to clipping. The simultaneous approach to the two lesions has been defended in different papers, both peritonal and supraorbital access being related.

The treatment of the aneurysm prior to the pituitary adenoma is in general the best option, be it by endovascular or microsurgical means. With the intention of reducing the risk, in view of the proximity of the aneurysm and the adenoma, the endovascular approach to the aneurysm followed by a transsphenoidal surgery to the adenoma has been preferred specially in unruptured aneurysms.

In patients with surgical pituitary adenomas, due to the routine pre-operative investigation with magnetic

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**Fig. 1** – (A–C) Cranial computed tomography (CT) showing subarachnoid hemorrhage with interhemispheric hematoma, associated with the suprasellar expansive lesion. In the CT performed 7 days following the ictus (D–F), a homogeneous contrast-capturing by the suprasellar lesion was observed.
Fig. 2 – Cerebral angiography showing anterior communicating complex aneurysm (A, B, D), associated with superior deviation of anterior cerebral arteries and narrowing of the left anterior cerebral artery (C). (A – right internal carotid artery/frontal incidence; B – right internal carotid artery/3D reconstruction; C – left internal carotid artery/frontal incidence; D – right internal carotid artery/3D reconstruction).

resonance, the angiography of cerebral vessels is not routinely requested. Nonetheless, according to Suzuki et al., in a study on a case of pituitary apoplexia caused by the rupture of an aneurysm in the internal carotid artery, cerebral angiography should be performed prior to surgery when the magnetic resonance imaging shows flow alterations, especially those in the anterior circulation.

In light of the above, it has been observed that the occurrence of cerebral aneurysms in patients with pituitary adenomas is larger than that in the general population and possesses a high potential for morbimortality. The factors which foster this association are not yet clear, but possible mechanisms are being discussed. We must therefore be attempt to suspect patients, as the correct diagnosis is crucial and may guide the therapeutic strategy, in genera in favor of prioritizing the aneurysm.

REFERENCES


