

Unruptured Internal Carotid Artery Aneurysm Associated with Functional Pituitary Adenoma: A True Association

Abstract

Detection of incidental intracranial aneurysm on neuroimaging can be associated with pituitary adenoma; however, such association is extremely rare. However, aneurysm with extension into the sella is extremely rare, and increases the risk of inadvertent intraoperative rupture, if trans-sphenoidal decompression is attempted either using endoscopic or microscopic surgical approach. Hanak *et al.* in a literature review of intrasellar noniatrogenic aneurysms over PubMed search in 2012 could only collect 31 studies, of which only eight cases had pituitary adenoma associated with aneurysm extending into sella. Authors report an interesting case of 52-year-old male diagnosed as case of acromegaly, was put on dopamine agonist, bromocriptine and responding well to therapy, however the magnetic resonance imaging raised suspicion of vascular pathology, and underwent digital subtraction angiography at our center revealed presence of right supraclinoid internal carotid aneurysm, which was coiled using endovascular technique with resultant good outcome.

Keywords: *Acromegaly, coexistence, pituitary adenoma, unruptured internal carotid aneurysm*

Introduction

Unruptured incidental intracranial aneurysm can co-exist with pituitary adenoma, however, occurrence is extremely rare, more so if aneurysm has extension into sellar cavity. Increased usage of neuro-imaging such as magnetic angiography and cerebral angiography for the screening of intracranial aneurysms in the recent years has led to increased detection of coexistence of intracranial aneurysms and brain tumors. However, the incidence of such coexistence is approximately 0.5% of all intracranial tumors as reported in an autopsy study by Housepian and Pool, but assessment of the real incidence rate is difficult, as angiography is not routinely performed for all brain neoplasms.^[1] Preoperative knowledge of asymptomatic aneurysms coexisting with pituitary adenoma can avoid accidental rupture of aneurysm during surgical resection of pituitary adenoma and/or may lead to planning of a special surgical strategy to deal with both pathologies simultaneously.^[2-5] Authors report a case of growth hormone (GH) secreting pituitary adenoma, which was associated with asymptomatic right supraclinoid internal carotid artery (ICA) aneurysm. Preoperative diagnosis of such coincidence requires a

high degree of suspicion and may seriously affect the functional outcome of the patient.

Case Report

A 52-year-old man presented with coarsening of facial features and insidious onset progressive decline in visual acuity for approximately 6 months duration. He consulted an endocrinologist, and underwent cranial magnetic resonance imaging (MRI) and diagnosis of GH secreting adenoma was made, accordingly kept on dopamine agonist, bromocriptine, which led to the symptomatic improvement of the acromegalic features, but the visual diminution continued to further worsening. After careful review of MRI, an associated vascular pathology in the right parasellar region was suspected. The patient underwent digital subtraction angiography (DSA) that showed right supraclinoid internal cerebral artery aneurysm [Figures 1-3]. On admission, in our department, the patient was alert and oriented. His visual acuity was 6/36 and 6/24 in the right and left eyes, respectively. Fundi examination revealed primary optic atrophy in the right eye. He had bitemporal hemianopsic visual field deficit on perimetry. Endocrinological evaluation revealed serum GH level of 6 ng/ml (prior to bromocriptine therapy 32 ng/ml), IGF1 level of 84ng/ml (prior to

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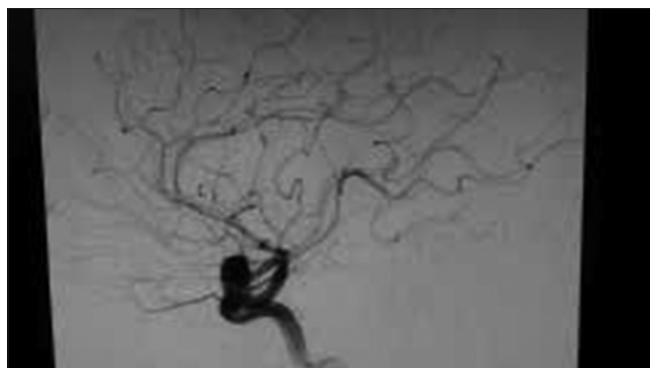


Figure 1: Cerebral digital subtraction angiography, lateral depicting showing right supraclinoidal internal carotid aneurysm in 55-year-old male



Figure 2: Cerebral digital subtraction angiography, anteroposterior views showing right supraclinoidal internal carotid aneurysm in 55-year-old male

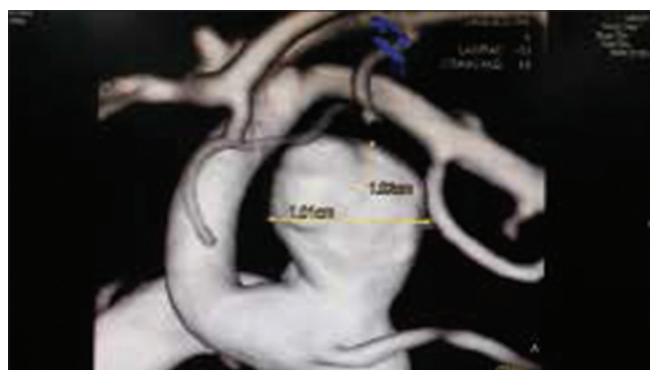


Figure 3: Cerebral digital subtraction angiography, three-dimensional reconstruction showing aneurysm of size 1.02 cm x 1.016 cm of right supraclinoidal internal carotid artery

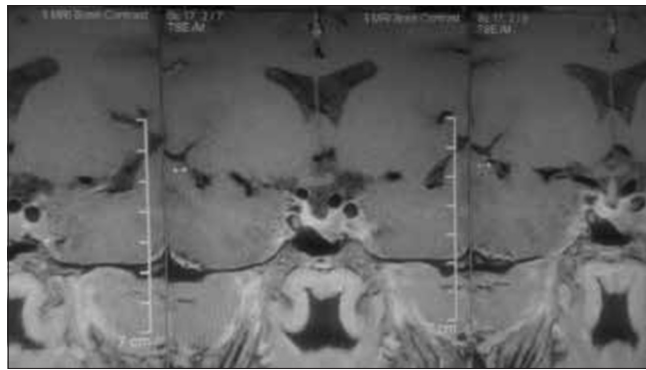


Figure 4: T1-weighted magnetic resonance imaging brain, coronal section showing sellar mass causing deviation of pituitary stalk

bromocriptine therapy, IGF1 625 ng/ml). Other pituitary hormone levels were all in the normal limits. MRI study revealed the presence of pituitary macro-adenoma with suprasellar extension with flow void in right parasellar region with extension into sella on T1 [Figure 4]. Cerebral angiography detected the presence of aneurysm located in the ophthalmic segment of the right ICA segment of the right ICA [Figures 1-3]. He was given the choice of surgery for management of pituitary adenoma decompression and clipping of aneurysm simultaneously using pterional craniotomy. In view of good response to dopamine agonist without adverse effect, he opted the medical management of adenoma and endovascular coiling of the aneurysm. His vision improved to 6/24 (right) and 6/18 (left) at 6 months after the coiling procedure.

Discussion

The association of intracranial tumors with asymptomatic aneurysms has been described in literature.^[4,5] In 1972, Pia *et al.*^[5] evaluated 116 intracranial tumor cases for coincidence with intracranial aneurysms. In this study, meningioma accounted for 29%, glioma 28%, and pituitary adenomas 21% of all brain tumors. Pituitary adenomas and meningiomas were associated with aneurysms twice as commonly as would have occurred from their independent occurrence. Isolated intracranial aneurysm

management itself presents a great surgical challenge and possess special catastrophic vascular risk if not diagnosed preoperatively.^[1,2,6-8] Furthermore, surgical risk can further escalate if the incidental aneurysm is associated with pituitary adenoma. Pituitary adenoma can be associated with intracranial aneurysms.^[2,3,9-16] Wakai *et al.*^[9] analyzed 95 cases of pituitary adenomas and reported the incidence of co-existing aneurysms was 7.4%, which was significantly higher than other brain tumors (1.1%). Jakubowski and Kendall^[10] reviewed angiographic studies on 150 surgically verified pituitary adenomas and observed co-existing aneurysm in 13.8% GH-secreting adenoma and 5.1% of nonsecretory adenoma and none with basophilic adenomas. In 2011, Manara *et al.*^[13] analyzed a total of 161 acromegaly cases, of which 152 cases were subjected for DSA evaluation of the circle of Willis. A total of 26 cases had 40 newly diagnosed induced aneurysms.^[5] In addition to two other patients, who previously undergone aneurysm clipping surgery.^[13] Intracranial tract of the ICA was the most common location.^[13] The association of intracranial aneurysms with GH level at the disease onset was associated with positive statistical correlation.^[13]

There are many explanations regarding high association of pituitary adenomas and intracranial aneurysms. Prolonged elevated GH level caused by GH-secreting adenoma may induce arteriosclerosis and degenerative changes in the arteries of the circle of Willis.^[9,11,17,18] Pia *et al.* speculated

local circulatory products secreted by tumors might have induced aneurysm. Even changes in the collagen metabolism can induce or even further promote aneurysm formation.^[5] Authors stated that tumor might stimulate aneurysm formation also by mechanical, microcirculatory, or other hormonal influences.^[17] Other postulates include the mechanical effect produced by microcirculatory changes in the tumor-supplying vessels.^[10] Mangiardi *et al.* advocated direct contact between the tumor and the aneurysm, with associated vascular infiltration.^[12] Housepian *et al.* believed that long-standing traction produced by pituitary adenoma adherent to the ICA wall can produce aneurysm.^[1]

Hanak *et al.* in a literature review over PubMed search could only collect 31 studies describing a total of 40 cases of intrasellar noniatrogenic aneurysms, being finally confirmed on MRI and angiography, of which only eight cases were associated with pituitary adenoma.^[19]

Pant *et al.*^[2] in a retrospectively reviewed 467 cases of pituitary adenomas, which all were subjected to DSA of at least anterior circulation reported that 25 (5.4%) cases of pituitary adenoma had intracranial aneurysms of which 97% were located in anterior circulation and 12% had multiple aneurysms. All of the aneurysm cases were incidental except two, who presented with bleeding. Incidence of aneurysms was more frequent with older age. Age distribution resembled that of aneurysm among general population. Although the association was most frequent among nonfunctional adenomas (8.8%) and least frequent among prolactinomas (2.4%). There was no statically significant association between hormone secreting status of pituitary adenoma, size, or invasiveness of tumor and presence of aneurysm. They suggested that such association is merely a chance factor, and the risk is not greater than that among the general population.

Parasellar aneurysms mimicking pituitary adenomas are usually asymptomatic, rarely present with hypopituitarism, pituitary apoplexy or rupture of aneurysm, epistaxis as a result of aneurysmal bleeding into the adenoma. Failure to anticipated possible presence of aneurysm and misdiagnosis of such coexistence can cause hazardous hemorrhagic complication if surgery is contemplated.

The presence of flow voids on T1-weighted and T2-weighted MRI sequences is 100% specific for aneurysms as described by Teng *et al.* with the sensitivity of 88%,^[6] however Olsen *et al.* reported that only 80% giant aneurysms show sign of blood flow in the aneurysm sac. Furthermore, MRI may fail to detect small sized intracranial aneurysms.^[7]

In the present case, axial and coronal MRI revealed a flow void in the right parasellar region, which was confirmed on cranial DSA.

Optimal management of such co-existing aneurysm with pituitary adenoma is controversial and depends on the size

of aneurysm, location of aneurysm in anterior or posterior circulation, proximity to sella or pituitary adenoma, size of pituitary adenoma, functional status of adenoma, size of adenoma, associated apoplexy manifestation, visual symptom, extension into multiple cranial fossa, or multi-compartmental of the tumor.

With increasing usage of endovascular approach, aneurysm can be coiled, and pituitary adenoma can be resected using microsurgical or endoscopic approach separately. However, aneurysms lying in close proximity to adenoma can be managed in a single stage with clipping of aneurysm and resection of the adenoma. Seda *et al.* reviewed the management strategy of an intracranial aneurysm associated with pituitary adenoma.^[14] The surgical approaches aimed for simultaneous treatment of the aneurysm and the pituitary adenoma through pterional or a supraorbital keyhole.^[14] Seda *et al.* managed their case with endovascular occlusion in the neck of ICA and transcranial clipping of ICA just below the ophthalmic artery's emergence. Moreover, pituitary adenoma was resected through a transsphenoidal route 1 month after the surgery of aneurysm.^[14] Seda *et al.* further advocated minimally invasive endovascular trapping of the aneurysms and medical treatment of GH-secreting pituitary adenoma.^[14]

Sade *et al.* reported a case of intrasellar aneurysm in a 39-year-old woman, DSA revealed presence of intracavernous carotid artery aneurysm which was managed with endovascular coiling.^[16] However, 8 months after coiling, she underwent transsphenoidal sub-total resection of pituitary adenoma. Sade *et al.* added a preoperative diagnosis of a coexisting intrasellar aneurysm in association with pituitary adenoma is an essential requirement to avoid life-threatening aneurysmal rupture and its consequences.^[16] Endovascular obliteration of intrasellar aneurysm represents an effective management carried out prior to attempting surgical intervention of pituitary adenoma.^[16]

High degree of suspicion for associated aneurysm is needed, and if MRI shows some atypical features, DSA must be carried out prior to contemplating surgical intervention to avoid iatrogenic aneurysmal rupture.

Conclusion

Asymptomatic aneurysm co-existing with pituitary adenoma constitute a rare but difficult to manage association, which has not received proper attention in the literature. Unawareness about coexisting aneurysm can produce catastrophic result if surgery is attempted. Preoperative knowledge can help to plan the therapeutic strategy either to deal with both lesions in a single operative satge or seperate management of the two pathologies successfully.

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Conflicts of interest

There are no conflicts of interest.

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