

Case Report

Enlargement of an incidental internal carotid artery aneurysm embedded in pituitary adenoma associated with medical shrinkage of the tumor: Case report

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Abstract

Background: Currently, transsphenoidal surgery (TSS) is the preferred method for surgical treatment of intrasellar pituitary adenomas. However, it carries some risk of intraoperative arterial injuries, which is mainly attributed to direct iatrogenic rupture of the internal carotid artery (ICA). There is anecdotal evidence suggesting that intracranial aneurysms are coincidentally found significantly more frequently in the setting of pituitary adenomas than when the incidence is compared to other intracranial neoplasms. The exact cause of this discrepancy remains unclear, but it certainly raises concerns about the potential existence of an ICA aneurysm, which might be encountered during TSS and in some cases may cause hemorrhagic complications.

Case Description: We present a case of a patient who was found to have a growth hormone (GH)-secreting pituitary adenoma and a coexisting cavernous ICA aneurysm which was embedded within the tumor. The patient underwent medical treatment of the adenoma. However, shrinkage of the tumor was associated with enlargement of the observed aneurysm, warranting endovascular intervention.

Conclusions: This case report is an illustration for physicians to be conscientious about the potential danger posed by the coexistence of an intratumoral aneurysm in the setting of a pituitary adenoma. Special attention should be given to recognition of an intrinsic flow void signal on the presurgical imaging of the tumor, and if observed, magnetic resonance angiography (MRA) should be performed for preoperative planning. If MRA is not performed routinely, detailed review of high-resolution magnetic resonance imaging is recommended to detect any flow artifacts suggestive of an aneurysm.

Key Words: Aneurysm, coincidence, complications, iatrogenic injury, intraoperative bleeding, pituitary adenoma, transsphenoidal

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INTRODUCTION

Currently, microscopic or endoscopic transsphenoidal surgery (TSS) is the preferred method for the treatment of intrasellar pituitary adenomas.^[13,17,19,32] Approximately 1,650 cases of TSS are performed annually for removal of pituitary adenomas in the United States.^[1,29] One of the preoperative considerations for TSS is an exclusion of “kissing internal carotid arteries: (ICA), a rare anatomical variant and absolute contraindication for TSS.^[25,28,38] However, even in the absence of this vascular configuration, the rate of intraoperative arterial injuries remains significant. In different case series, it has been reported to be as high as 1.1%.^[2,8,9,14] Thus, the imputed number of iatrogenic vascular injuries encountered during TSS in the United States is approximately 18 cases per year, which is a significant cause of morbidity and mortality as well as physician liability. These injuries are mainly attributed to direct iatrogenic injury to the ICA.^[3,18,25,39] However, there is some evidence suggesting that coincidental intracranial aneurysms are found almost seven times more frequently in pituitary adenomas when compared to other intracranial neoplasms.^[6] The exact cause of this augmented incidence still remains unclear; however, it raises concern about encountering an ICA aneurysm during TSS, which in some cases may lead to hemorrhagic complications.

We present a case of a patient who was found to have a growth hormone (GH)-secreting pituitary adenoma and a coexisting cavernous ICA aneurysm embedded within the tumor. The patient underwent medical treatment of the adenoma. However, shrinkage of the tumor resulted in simultaneous enlargement of the observed aneurysm warranting endovascular intervention.

CASE REPORT

Initial management

A 37-year-old female presented to our department after noticing progressive enlargement of her jaw, ears, nose, and fingers. Physical examination and comparison to previous photographs confirmed acromegaly. Further neurological examination including formal visual field testing was within normal limits. A full spectrum of pituitary axis investigations was performed, which revealed increased levels of IGF-1 (1109 ng/ml) as well as increased levels of GH (15 ng/ml). All other hormonal laboratory results were within the normal reference range.

High-resolution magnetic resonance imaging (MRI) of the brain with contrast media was performed, which revealed a 17.5 mm × 15.7 mm × 17.4 mm pituitary macroadenoma with stalk displacement. The patient was admitted to our department for elective transsphenoidal surgery. During the preoperative review of MRI films,

a subtle area of flow void was noticed within the right anterior aspect of the tumor [Figure 1].

The decision was made to perform a magnetic resonance angiography (MRA) for a more detailed visualization of the vasculature, which revealed a 2.5-mm cavernous ICA aneurysm pointing towards the sella.

A detailed discussion was then carried out with the patient about the natural history of adenomas and all available treatment options, including endovascular treatment of the aneurysm followed by TSS. We also discussed associated risks and benefits as well as the risks of leaving the adenoma untreated. The patient elected to first pursue medical treatment for pituitary adenoma with close follow-up for the aneurysm.

Continued management

Shortly after initiation of monthly intramuscular injections of 30 mg of octreotide depot and twice a week administration of 0.25 mg oral cabergoline, the patient noticed a significant decrease in the soft tissue swelling of her face and extremities. IGF-1 levels showed a steady decline from 1109 ng/ml to 353.1 ng/ml over the follow-up period of 1.5 years. The efficacy of medical therapy was also confirmed by a steady decline in tumor size from 17.5 mm × 15.7 mm × 17.4 mm to 10.9 mm × 4.9 mm × 8.9 mm [Figure 2a-c]. Simultaneously, there was a gradual increase in the aneurysm size from 2.7 mm to 4 mm [Figure 2d-f].

Six months later a CT angiography was performed at an outside hospital where the patient was seen for a second opinion [Figure 3].

Given the steady increase in the size of the aneurysm and considering the absence of reliable data about the natural history of intratumoral aneurysms, the patient was again offered endovascular treatment and subsequently underwent a successful stent-assisted coil embolization

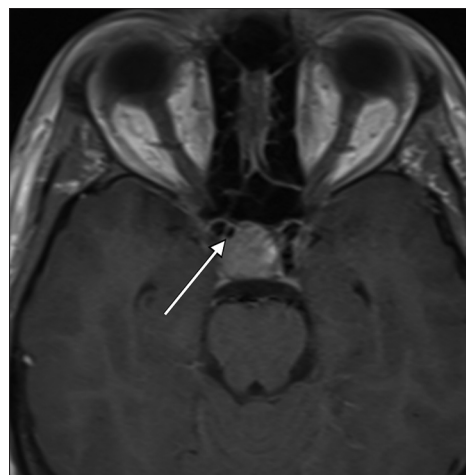


Figure 1: Contrast-enhanced MRI of the pituitary region showing a subtle area of flow void (arrow) in the right anterior aspect of pituitary adenoma

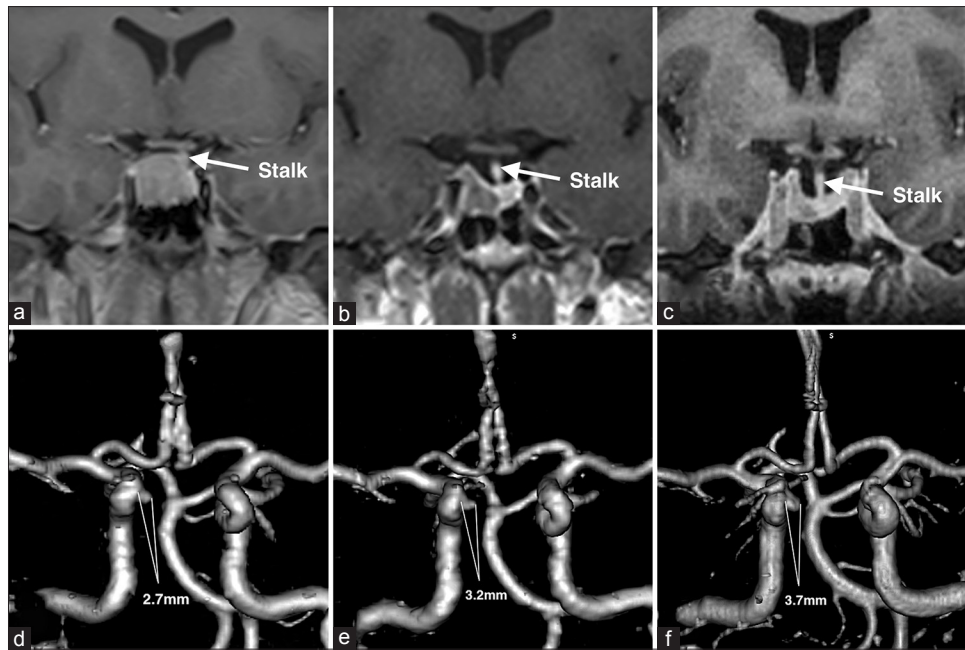


Figure 2: Contrast-enhanced MRI of the pituitary region demonstrating gradual decrease in tumor size over 18 months follow-up and returning of pituitary stalk to its physiologic position (white arrows), (a-c) as well as corresponding MRA follow-up demonstrating enlargement of ICA aneurysm (d-f)

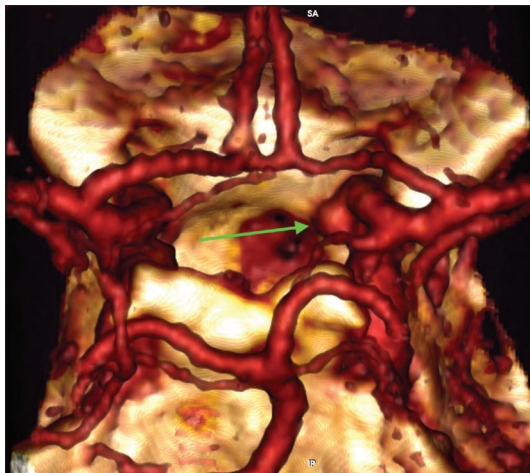


Figure 3: CTA of the circle of Willis demonstrating a 4.5 mm × 4 mm aneurysm of the right cavernous ICA (arrow)

of the aneurysm without complications [Figure 4]. The stent was placed due to the relative low dome-width to neck-width ratio as well as to secure the coil in place in case subsequent TSS was required. One-year follow-up angiogram revealed complete occlusion of the aneurysm along with continuous decrease of IGF-1 levels [Figure 5]. Currently, the patient remains asymptomatic and we shall follow-up with clinical, radiographic, and laboratory surveillance at regular intervals.

DISCUSSION

Rupture of aneurysms located in the parasellar cavernous segment of the ICA occurs rarely. It is usually clinically

silent though cranial nerve deficits have been reported in some cases.^[11,20,34,35] To our knowledge, there are only two reported cases of ICA aneurysms embedded within a pituitary adenoma.^[6,26] On the other hand, intraoperative iatrogenic injury to the ICA is a known and much feared complication with a rate of as high as 1.1%.^[2,8,9] However, there is only one case report of an iatrogenic rupture of an aneurysm embedded within the pituitary adenoma.^[27] Given the fact that routine preoperative MRA investigation are not advocated in the guidelines of management of pituitary adenomas,^[5] it can be assumed that some of these injuries are caused by the rupture of an associated aneurysm.^[17,21,25]

TSS is the preferred method of treatment for GH-secreting tumors, and medical therapy is indicated in cases when contraindications to surgery exist.^[21,29] Among other relevant workup, standard preoperative planning of TSS includes review of the distinct anatomical characteristics of the individuals' sellar and sphenoid region as well as the course of the internal carotid arteries.^[4, 28] Most often, physicians spend only seconds on the examination of printed MRI films to exclude the presence of “kissing” carotids; this coupled with any other eye-catching pathology on films, such as a pituitary macroadenoma, may draw the attention of radiologists away from subtle flow artifacts. Thus, there is some concern that an incidental aneurysm may be present within the sellar region and become the cause for catastrophic intraoperative complications.^[26]

There are several case reports about the development of a postoperative pseudoaneurysm of the ICA.^[22-24,37]

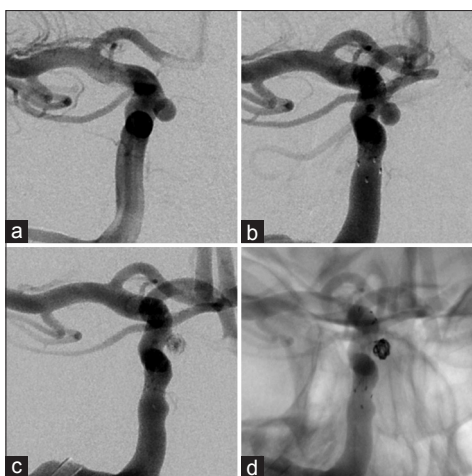


Figure 4: Stent-assisted coil embolization of cavernous ICA aneurysm: (a) Preoperative angiogram. (b) Angiogram following the placement of the stent. (c and d) Postoperative angiograms with and without subtraction

Detailed review of these cases revealed that these patients were not evaluated for the presence of an aneurysm prior to surgery.^[22-24,37] Thus, it is difficult to conclude whether there was a preexisting aneurysm as opposed to a newly-formed pseudoaneurysm.

According to Choi *et al.*, incidental intracranial aneurysms are found seven times more frequently in pituitary adenomas than in other types of tumors.^[6] The underlying pathological mechanism for the observed higher incidence of intracranial aneurysms in GH-secreting pituitary adenomas (described by Spitler *et al.*) fails to provide a sufficient explanation for a higher incidence in other types of pituitary adenomas.^[31] Thus, the exact cause of this peculiar peak in incidence remains unclear; however, it certainly raises concerns about the potential coexistence of an ICA aneurysm in the setting of an adenoma which may lead to hemorrhagic complications such as subarachnoid hemorrhage, carotid cavernous fistula, pituitary apoplexy, and intraoperative massive bleeding.^[3,7,10,12,15,16,30,39] Therefore, a detailed assessment and review of the cerebral vasculature on presurgical MRI scans is required.

Finally, there is no reliable data about the natural history of coexisting aneurysms in the presence of GH-secreting pituitary adenomas. Spitler *et al.* have found a higher incidence in aneurysm growth rate in patients suffering from GH-secreting tumors.^[31] Assuming that the presence of these pathologies is a pure coincidence, and thus, managing it as an isolated condition is in contradiction to the aforementioned statements. However, in the absence of sufficient data and a scarcity of such reports in the literature, guideline-based management of incidentally discovered aneurysms should be considered.^[36] The currently established guidelines for the management of unruptured intracranial aneurysms reflect the

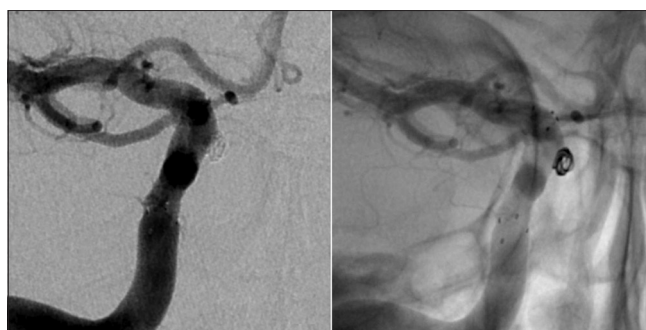


Figure 5: One-year follow-up diagnostic angiography demonstrating complete occlusion of the aneurysm with digital subtraction (left) and without digital subtraction (right)

propensity for growing aneurysms to rupture and warrant endovascular treatment of unruptured intracranial aneurysms in this setting.^[33]

CONCLUSIONS

Single case reports do not provide enough evidence to draw scientific conclusions. However, they can serve as a reminder about potential perils in daily practice.

- Eye-catching pathologies often draw attention away from other subtle coincidental findings. Neurosurgeons must remain alert about possible coincidental pathologies, which may lead to devastating intraoperative complications.
- Special attention should be devoted to recognition of a flow void within the tumor and MRA should preferably be performed during the preoperative planning. If MRA is not performed, detailed review of high-resolution MRI should be conducted to reveal any flow artifacts suggestive of an aneurysm.
- The association of tumor shrinkage and aneurysm enlargement in our case does not imply causality. A prospective multicenter screening study could reveal the true incidence of intrasellar aneurysms associated with pituitary adenomas and compare its natural history to that of other types of intracranial aneurysms.

Patient consent

The patient has consented to submission of this case report to the journal.

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Nil.

Conflicts of interest

All authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest (such as honoraria; educational grants; participation in speakers' bureaus; membership, employment, consultancies, stock ownership, or other equity interest; and expert testimony or patent-licensing

arrangements), or non-financial interest (such as personal or professional relationships, affiliations, knowledge or beliefs) in the subject matter or materials discussed in this manuscript.

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