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Case Report

Management dilemma in a rare case of pituitary apoplexy with akinetic mutism in the setting of ruptured junctional brain aneurysm: A case report and literature review

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ABSTRACT

Backgound: Pituitary apoplexy is associated with stroke, head injury, and brain tumors. Still, its presentation due to the ruptured aneurysm is rare and its presentation with akinetic mutism has not been reported.

Case Description: The patient in the present study is 21-year-old female who presented in our emergency department in an altered sensorium with Glasgow comma score (GCS) E2V1M1. She was intubated and resuscitated. Routine blood investigations, lipid profile, and hormonal studies were normal. Initial noncontrast computed tomography (NCCT) head revealed subarachnoid hemorrhage in the interhemispheric fissure and evidence of bleeding in the pituitary gland. Magnetic resonance imaging (MRI) brain was soon done, which showed an infarct and hemorrhage in the pituitary gland; there was an evidence of an infarct in the bilateral medial frontal gyrus, basal ganglia, and supplementary motor area. MR arteriography revealed an aneurysm at the left A1-anterior communicating artery (Acom) junction directed superomedially with diffuse spasm in a bilateral anterior cerebral artery. Pterional craniotomy was done with clipping of the aneurysm and evacuation of blood clots from the interhemispheric fissure and pituitary gland. Histopathology features suggestive of the non-functioning pituitary tumor with interspersed hemorrhagic necrosis. Intraarterial vasodilation with microcatheter injection was given, but vasospasm did not improve. Postoperatively, Levodopa was started. She used to track objects in front of her eye and started nodding her head in "yes and no fashion," with power in limbs improved to 3/5 at 6 months of follow-up.

Conclusion: Pituitary apoplexy with ruptured A1-Acom junction aneurysm with nonfunctioning pituitary macroadenoma is rare, and its presentation with akinetic mutism has not been reported. As there is scarce literature suggesting an association between pituitary apoplexy and ruptured aneurysm, it is challenging to comment regarding its pathogenesis. Although akinetic mutism generally has a poor prognosis, it may respond to Levodopa with a better outcome.

Keywords: Akinetic mutism, Pituitary apoplexy, Ruptured brain aneurysm, Ruptured intracranial Aneurysm

INTRODUCTION

Pituitary apoplexy is defined as a hemorrhage or infarct in the pituitary gland. Pituitary apoplexy due to a ruptured anterior circulation aneurysm is uncommon and rare due to an A1 segment of the anterior cerebral artery (ACA) aneurysm.[12] Association of akinetic mutism with ruptured

anterior circulation aneurysms is also rare and very few cases have been reported.[1,2,8] Such cases are challenges for diagnosis and management. In this case report, we reviewed the etiology, the association between pituitary apoplexy and ruptured intracranial aneurysm formation, and the course of recovery of the patient following treatment.

PATIENT DETAILS

A 20-year-old female patient presented with altered sensorium, and quadriparesis, unable to speak for 7 days. Cranial nerve function, including 2nd, 3rd, 4th, and 6th, could not be assessed. Power in bilateral upper and lower limbs was 1/5. Blood investigations were normal, including hemoglobin (HB), total leucocyte count, differential leucocyte count (TLC, DLC), electrolytes, liver function test, and coagulation profile. Preoperative hormonal assays were normal and viral markers were negative. On radiological evaluation, the initial NCCT head revealed a subarachnoid hemorrhage in the interhemispheric fissure and evidence of bleeding in the pituitary gland [Figure 1a]. MRI brain was soon done, which showed an infarct and hemorrhage in the pituitary gland; there was an evidence of an infarct in the bilateral medial frontal gyrus, basal ganglia, and supplementary motor area [Figure 1b]. MR arteriography revealed an aneurysm at the left A1-anterior communicating artery (Acom) junction directed superomedially with diffuse spasm in a bilateral anterior cerebral artery (ACA) [Figure 1c]. The patient was started on oral nimodipine and other medications for symptomatic treatment. The left pterional craniotomy with clipping of aneurysm and evacuation of hematoma in the pituitary gland, gyrus rectus, and interhemispheric fissure was done together with excision of pituitary macroadenoma. Postoperative digital subtraction angiography was done to assess for aneurysm occlusion and collateral flow. It revealed non-visualization of the aneurysm with complete occlusion of the aneurysm neck and vasospasm in the bilateral anterior cerebral and middle cerebral arteries (MCAs). Intraarterial spasmolysis by microcatheter in the bilateral internal carotid artery (ICA) was tried without much improvement in vascular spasm [Figures 2a-d].

The postoperative MRI brain revealed a bilateral diffuse infarct [Figure 3a]. Postoperative histopathology revealed blood clots intermixed with tumor tissue on hematoxylin and eosin (H&E) staining and no staining on immunohistochemistry for prolactin, growth hormone, and adreno corticotrophic hormone (ACTH) [Figures 3b-d]. Postoperatively, there was an improvement in

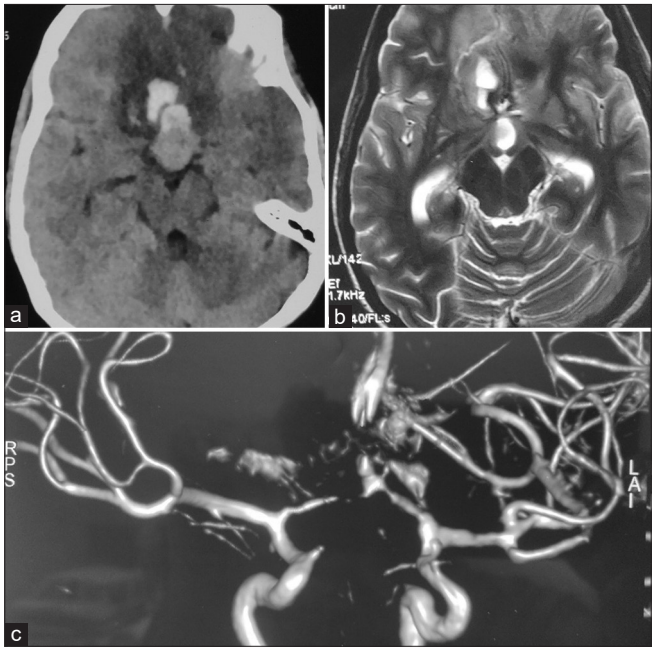

Figure 1: (a) Non-contrast computed topography head suggests hemorrhage in the pituitary gland and interhemispheric fissure with hypodensities in the bilateral medial frontal lobe suggesting infarct; (b) T2 axial MRI suggests a hyperintense lesion in the isointense pituitary gland and hyperintensity in the medial frontal lobe with bilateral loss of sulci and gyri pattern suggestive of the infarct; (c) MR arteriography suggests an aneurysm at the left A1-Acom junction with no visualization of proximal and distal anterior cerebral artery suggestive of diffuse spasm. RPS: Right posterosuperior; LAI: Left Anteroinferior.

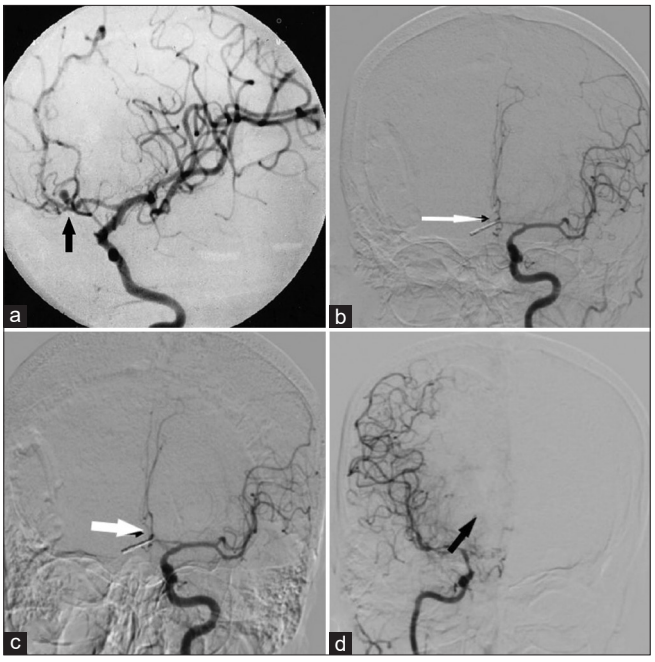

Figure 2: (a) Preoperative digital subtraction angiography (DSA) showing anterior communicating artery (Acom) aneurysm at the junction of the Acom and left A2 with distal vessels narrow in caliber, suggesting vasospasm (black arrow); (b) postoperative DSA showing clipped aneurysm with diffuse vasospasm of bilateral A2, Left A1, and middle cerebral artery vessels (white arrow); (c) postoperative DSA following vasodilation by intraarterial microcatheter injection of nimodipine revealing persistent vasospasm (white arrow); (d) postoperative DSA of the right internal carotid artery suggestive of persistent vasospasm more in anterior cerebral artery branches than right middle cerebral artery (black arrow).

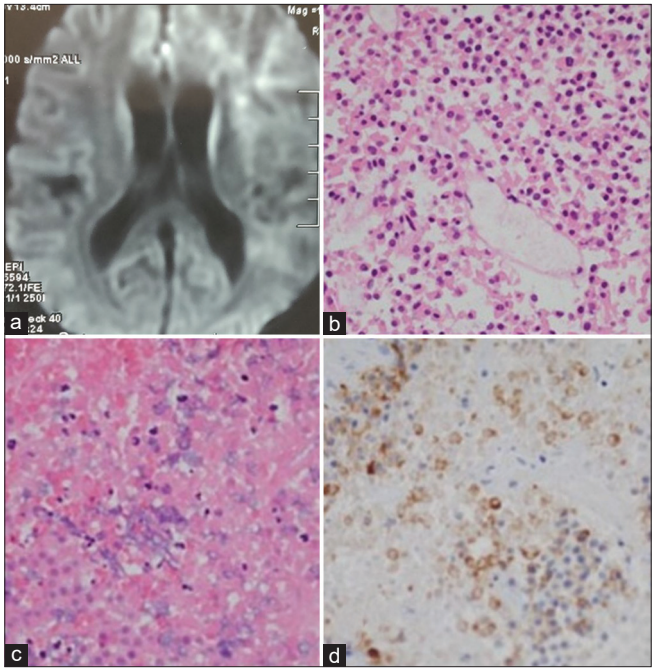

Figure 3: (a) Postoperative MRI brain with diffusion-weighted sequence suggests restriction diffusion in the bilateral frontal lobe, and basal ganglia region suggestive of the infarct; (b) microscopic sections show a hypercellular tumor composed of solid sheets, nests, and trabeculae. Tumor cells contain scant granular eosinophilic cytoplasm, round nuclei inconspicuous nucleoli; (c) Hematoxylin-Eosin staining of the pituitary macroadenoma revealing hemorrhagic necrosis pattern and extensive tumor coagulative necrosis, consistent with recent hemorrhagic infarction; (d) immunohistochemical results for pituitary hormones show negative staining for growth hormone, leutinising hormone, thyroid stimulating hormone, adreno corticotrophic hormone (GH, LH, TSH, and ACTH); Prolactin immunostains suggest nonfunctional pituitary macroadenoma.

vision that can be assessed by the fact that the patient followed the objects, showed visual gestures on commands, and talked on the mobile phone. Her limb power remains to be 1/5. She was started on Levodopa. She has slow improvement in the power of her limbs after 6 months of follow-up to 3/5. Physiotherapy and nursing care are being continued at home.

DISCUSSION

The intracranial aneurysm has been reported earlier, with a higher prevalence in pituitary adenoma than in the general population.[2] Secretary pituitary adenomas are more commonly associated with aneurysms than nonfunctioning pituitary adenomas. The prevalence of unruptured aneurysm with growth hormone-secreting adenoma is 2.3–13% and with prolactin-secreting adenoma is 0.9–9%.[6,10] In the literature, most pituitary apoplexy cases reported with ruptured aneurysms were found to have growth hormone-secreting tumors followed by prolactin-secreting, and probably one patient is reported to have nonfunctioning pituitary macroadenoma similar to the present patient.[12]

As reported earlier by a few authors, the secretary nature of the tumor may induce weakness in the wall of the vessels close to it.[6] It has been reported that tumors had an extension to the cavernous sinus and parasellar extension may induce changes in the wall of vessels close to the tumor.[6,10]

In the present case, the tumor was neither functional nor had parasellar extension to support the hypothesis proposed in the earlier studies that pituitary macroadenoma could have influenced the formation of the aneurysm by manipulating the course of the vessels or have influenced the rupture by secretary nature of the tumor. In this case, the aneurysm was directed superomedially at the junction of the left A1 and Acom. The lower lobe of the aneurysm had ruptured, leading to hemorrhage in the pituitary tumor and bleeding in the interhemispheric fissure. Such findings in the patient had been reported by Yoshida *et al*. in non-functioning pituitary macroadenoma and Laidlaw *et al*. in growth hormone-secreting tumors. Still, none of them have features suggestive of akinetic mutism.[4,12] In the earlier reported five patients with similar ruptured aneurysm and pituitary apoplexy without akinetic mutism, one had prolactinoma, two had growth hormone-secreting, and two had hormone profile which was not suggestive of secretary tumor.[3-5,7,9,11,12]

The patient in the present study was 21-years-old female; the youngest patient reported pituitary apoplexy due to a ruptured aneurysm. Earlier, Song *et al*. reported a patient 31 years of age with a left Pcom aneurysm with unknown pituitary secretary function, and clinically, the patient had headaches, vomiting, and visual blurring.[9]

Preoperative severe vasospasm in the distribution of bilateral distal ACA and MCA territory may have led to infarct in the region of bilateral supplementary motor area, thalamus, hypothalamus, and basal ganglia leading to akinetic mutism in this patient. Akinetic mutism is generally observed following head injury, stroke, and brain tumors. Still, its presence with a ruptured A1-Acom junctional aneurysm is rare, and its association with pituitary apoplexy probably had not been reported to the best of my knowledge.

In a literature review by Choudhari *et al*., 21 cases have been reported with akinetic mutism following a ruptured aneurysm with subarachnoid hemorrhage. In the majority of the patients, they have infarct in distal ACA territory with poor prognosis.[2] In the present patient, patients had been started with Levodopa postoperatively. She had improved vision, probably due to the evacuation of hematoma in the pituitary gland and the interhemispheric fissure. She followed the objects in front of her eye and responded to commands in the form of opening and closing her eyes and head nodding to reply yes or no. Patients have mild improvements in limb

movement on 6-month follow-up, probably due to Levodopa. In such a scenario, Levodopa had been reported to respond only in rare selected patients, as Combarros *et al*.[3] In the study by Sibille *et al*., better recovery was reported in patients with akinetic mutism following the evacuation of hematoma around the cingulate gyrus and less extensive infarct in ACA territory, contrary to the present case.[8]

CONCLUSION

It is difficult to comment on the exact etiology of the prevalence of aneurysm with pituitary macroadenoma, especially the growth hormone-secreting prolactinoma, as it can also present in nonfunctioning pituitary macroadenoma. Presentation with akinetic mutism with infarct in the distal territory is rare, with pituitary apoplexy with ruptured A1-Acom junctional aneurysm. Improvement following levodopa supplementation has not been encouraging in distal ACA territory infarct with akinetic mutism. Still, its role in improvement cannot be discarded as found in the present case so that it can be given in such a scenario.

Declaration of patient consent

Patient's consent not required as patient's identity is not disclosed or compromised.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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