Case report

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A case of pituitary apoplexy with unusual presentation

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Abstract

Pituitary apoplexy is a rare but potentially life-threatening condition caused by either haemorrhage or infarction of the pituitary gland. It commonly occurs in non-functioning pituitary adenoma. Patients present with the clinical syndrome of headache, visual defects or ophthalmoplegia, altered consciousness and variable endocrine deficits. However, the condition may also be subclinical, like in our case, where the presenting feature was mild to moderate headache for a few months, misguiding the physician. Her prolactin level was high, and an MRI revealed evidence of haemorrhage within the pituitary gland. Her amenorrhoea persisted even after normalization of prolactin, which may be related to reduced LH and FSH after apoplexy. The case is reported because of its simplicity of presentation, which we should not ignore. *[J Assoc Clin Endocrinol Diabetol Bangladesh,* July 2024;3(2)*: 67-70]*

Keywords: Pituitary adenoma, Pituitary apoplexy, Pituitary infarction, Pituitary surgery

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Introduction

The pituitary is a tiny 'master endocrine gland' located at the base of the brain and is connected to the hypothalamus. Any of the pituitary hormones may be affected in pituitary disease, with acute adrenocorticotropic hormone (ACTH) deficiency being the most catastrophic and life-threatening. Pituitary apoplexy occurs following acute haemorrhage or infarction of the pituitary gland, causing patients to be acutely unwell due to hormonal as well as local compressive effects.[1] These effects cause the usual presentation of pituitary apoplexy, such as severe headache, diplopia, visual loss, and hypopituitarism.[2] However, the severity of presenting symptoms may vary.

Case report

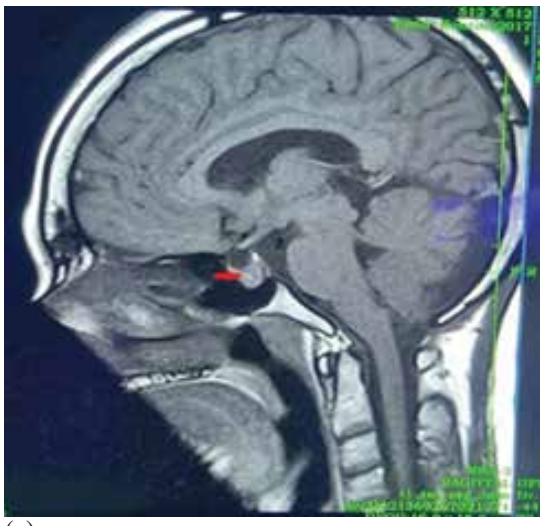
A 28-year-old Bangladeshi female visited the outpatient department of our hospital with complaints of headaches and amenorrhea for the last 3 months. The headache was chronic, mild to moderate in intensity, global, without any definite aggravating or relieving factors. Before 3 months, her menstrual cycle was regular. She had two children; the last child was eight years old. She had no history of galactorrhea. There was no history of visual disturbances, vomiting, seizures, or unconsciousness. She had not taken any medications except analgesics like paracetamol.

On clinical examination, she was fully conscious and oriented. Her body mass index (BMI) was 23.4 kg/m^2, blood pressure 110/70 mmHg without any postural drop, pulse rate 100 beats/min, body temperature 98^{o}F, and oxygen saturation 100%. All the cranial nerves were intact and there was no papilledema. An ultrasonogram of the abdomen revealed normal uterus and adnexa.

Endocrine workups were remarkable for high prolactin and low LH and FSH (Table-I). Biochemical tests were unremarkable. Magnetic resonance imaging (MRI) with contrast study revealed a small hemorrhagic focus

Table-I: Laboratory findings of the patient

Test	Initial result	After treatment	Normal value
Prolactin (ng/mL)	313.2	29.8	3.0-16.6
Free T4 (ng/dL)	1.10	-	0.9-2.3
TSH (mIU/L)	3.0	-	0.4-4.0
Morning cortisol (mcg/dL)	14	-	3.7-19.0
LH (mIU/ml)	0.31	-	1.7-15
FSH (mIU/ml)	1.54	-	1.4-9.9



(a)

(b)

(c)

Figure-1: MRI of the brain showing small hemorrhagic focus within the anterior lobe of the pituitary gland; (a) T1 weighted image sagittal view, (b) T2 weighted image sagittal view, (c) T2 weighted image coronal view

within the anterior lobe of the pituitary gland which represents a possible resolving apoplexy in the background of a prolactinoma (Figure-1).

She was treated with cabergoline 0.50 mg 2 tablets twice a week. She tolerated the drugs well without any remarkable side effects. After 3 months, her prolactin level was 29.8 ng/ml, and the dose was reduced to 1 tab twice a week. Subsequent follow-up after 3 months revealed prolactin level 15 ng/ml. Other hormonal profiles were normal. As she was experiencing amenorrhea, hormone replacement therapy (HRT) was started along with a reduced dose of cabergoline. Unfortunately, the patient was lost to follow-up.

Discussion

The first case of a pituitary tumour associated with haemorrhage was described by Bailey in 1898.[3] The signs and symptoms of pituitary tumour infarction or haemorrhage were collectively coined as 'pituitary apoplexy' by Brougham et al. in 1950.[4]

Pituitary apoplexy is a rare condition secondary to abrupt haemorrhage or infarction, which complicates 2-12% of pituitary adenomas, especially non-functioning tumors.[5] The clinical presentation of pituitary apoplexy is highly variable and is largely determined by the extent of hemorrhage, necrosis, and edema. Local compression of adjacent structures may cause sudden onset of excruciating headaches, visual disturbances (cranial nerve palsies and ophthalmoplegia), and other symptoms of raised intracranial pressure.[6] Headache is present in more than 80% of the patients with pituitary apoplexy and is generally the initial manifestation.[1,7] Headache occurs secondary to meningeal irritation due to blood entering the basal cisterns or dural stretching. Blood in the supra-sellar cistern may provoke chemical meningitis. Restricted visual fields result from distension of the optic nerves and chiasma, specifically bitemporal hemianopia. Lateral haemorrhage and necrosis lead to deficits in the 3rd, 4th, 5th, and 6th cranial nerves.[8]

Pituitary apoplexy may also be associated with life-threatening pituitary insufficiency or an acute adrenal crisis. Clinically, the most crucial deficit is ACTH, which has been reported in up to 70% of the patients. Thyrotrophin and gonadotrophin deficiencies are observed in 50% and 75% of the patients. Hyponatremia has been reported in up to 40% of the patients because of either SIADH or hypocortisolism. Patients with pituitary apoplexy who have low serum prolactin levels at presentation have the highest

intrasellar pressure and are the least likely to recover from hypopituitarism after decompressive surgery.[9] A decreased level of consciousness may result from endocrine abnormalities associated with hypopituitarism or pressure transmitted to the brainstem or owing to hypothalamic compression.[10] Diabetes insipidus is rarely a feature because the posterior lobe is usually unaffected.

Commonly, the episode of apoplexy occurs spontaneously; however, several precipitating factors have been identified in up to 40% of cases of pituitary apoplexy. Hypertension was by far the commonest (26%) predisposing factor; others are coronary artery bypass grafting (CABG), anticoagulation, dynamic pituitary function tests, radiation, bromocriptine therapy for existing macro-adenoma, trauma, surgery, pregnancy, elevated oestrogen levels and diabetes.[11] No precipitating factors could be identified in more than 50% of pituitary apoplexy cases.[1] However, some cases (up to 20%) of pituitary apoplexy are subclinical, only revealed by MRI performed during follow-up of known pituitary adenoma.[12]

Pituitary apoplexy can be difficult to diagnose clinically. Computed tomography (CT) is the most useful in the acute setting (24-48 h). However, a CT scan cannot detect all cases of pituitary apoplexy, especially if CT is obtained after the first 48 hours of the onset of haemorrhage. MRI is useful for identifying blood in the subacute setting (4 days to 1 month). It also helps to exclude other acute cerebrovascular events, specifically subarachnoid hemorrhage, bacterial meningitis, cavernous sinus thrombosis, and intrasellar mass.[10,13]

The first intervention after pituitary apoplexy diagnosis is hemodynamic stabilization, correction of electrolyte disturbances, and corticosteroid administration. All patients should have urgent blood samples for biochemistry and hormonal assessment (random cortisol, prolactin, FT4, TSH, IGF1, GH, LH, FSH and testosterone in men, oestradiol in women). After collecting blood samples, glucocorticoids should be administered immediately after onset in supraphysiological doses to serve as a replacement for endogenous hormone deficiency and help control the effect of edema. In adults, hydrocortisone 100-200 mg IV/IM bolus followed by either 2-4 mg/hr by continuous IV infusion or 100 mg iv 6 hourly. Dexamethasone is not favoured as glucocorticoid replacement, although it may reduce oedema as a nonsurgical strategy for treating pituitary tumour apoplexy.[14] For patients with lactotroph adenoma, administration of dopamine agonists can reduce adenoma size.

Owing to the highly variable course of this syndrome and limited clinical experience, the surgical management of pituitary apoplexy is controversial. Some authors advocate a conservative approach for selected patients (those without visual acuity or field defect), while others adopt surgical decompression for all patients with pituitary apoplexy. Conservative management is increasingly used in selected patients, yielding similar outcomes. Pituitary apoplexy is a neurosurgical emergency that may require urgent surgical decompression to prevent permanent visual loss and possible death from local pressure on the hypothalamus and brainstem.[1]

The case discussed here illustrates an unusual clinical presentation of only mild to moderate chronic headache and amenorrhoea. It is important to recognize these mild presentations so that definitive therapy can be instituted. If undetected, life-threatening, catastrophic incidences can occur from adrenal crisis. Her amenorrhoea persisted even after normalization of prolactin, which may be related to reduced LH and FSH after apoplexy. However, we could not see the changes over long-term follow-up, nor could we examine the repeat MRI, which was essential in this case.

Conclusion

Pituitary apoplexy can present in various clinical pictures, from mild symptoms, such as in our patient, to severe symptoms. It should be considered in a patient with headaches and endocrine dysfunctions. MRI is the most sensitive diagnostic imaging modality, and prompt diagnosis expedites immediate treatment. A high index of suspicion is required even in mild symptomatic cases for early diagnosis and appropriate treatment of this potentially fatal disease.

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Disclosure

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Data Availability

Any inquiries regarding supporting data availability of this study should be directed to the corresponding author and are available from the corresponding author on reasonable request.

Ethical Approval and Consent to Participate

Written informed consent was obtained from the patient. All methods were performed in accordance with the relevant guidelines and regulations.

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