

9. Wortsman J, Hughes LF. Case report: olfactory function in a fertile eunuch with Kallmann syndrome. *Am J Med Sci.* 1996;311:135–8.

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Silent pituitary infarction of an uncommon etiology[☆]



Infarto hipofisario silente de etiología infrecuente

Pituitary apoplexy (PA) is an uncommon, life-threatening clinical syndrome that occurs after acute pituitary hemorrhage or infarction. PA may occur in a pituitary gland with no prior pathological process, but usually occurs as a complication of an adenoma.¹ Its clinical presentation varies widely, and may consist of a nonspecific clinical picture, typical signs and symptoms (headache, nausea, vomiting, visual changes) or, in most severe cases, pituitary insufficiency leading to coma and death. Subclinical presentation is most uncommon. This, combined with the low prevalence of the syndrome, makes diagnosis difficult, with the resultant increase in morbidity and mortality. We report the case of a patient with no relevant personal history who experienced PA secondary to tooth extraction, an etiology not previously reported.

A 41-year-old male was referred to our endocrinology outpatient clinic by his family physician for hypotension and chronic anemia. The patient had an unremarkable family and personal history, except for tooth extraction four years before with bleeding of approximately 2 L that required hospital admission and treatment with plasma expanders. The patient had had since then a nonspecific clinical picture consisting of anorexia and weight loss, abdominal discomfort, intolerance to cold, normocytic normochromic anemia, and decreased libido without impotence. Physical examination found low blood pressure levels (90/60 mmHg), normal weight (height 172 cm and weight 68.7 kg), no goiter on palpation, and dry and rough skin. No abnormal findings were made in cardiopulmonary, abdominal, or limb examination. Hypopituitarism was suspected, and baseline hormone tests were therefore performed with the following results: basal serum cortisol (1st and 2nd measurements): 2.3 and 3.15 g/dL (normal: 5–30), ACTH 6.7 pg/mL (10–80), prolactin 6.8 ng/mL (2.5–7.5), TSH 0.15 U/mL (0.30–5.5), FT4 3.9 pg/mL (8.5–18), LH 6.95 mU/mL (0.6–12), FSH 3 mU/mL (1.0–8.0), and total testosterone 3.92 ng/mL (1.8–18.5). Based on a diagnosis of corticosteroid and thyroid deficiency, replacement therapy was started with hydrocortisone and

levothyroxine. To diagnose a potential GH deficiency, the insulin-induced hypoglycemia test was performed: glucose 88, 39, 30, and 20 mg/dL (the test was discontinued at this blood glucose level); GH 0.19, 0.49, 2.78, and 2.84 ng/mL respectively. Basal IGF-1 was 65 ng/mL (normal: 90–360). After checking patient compliance with protocol criteria, treatment for such deficiency was started.

Magnetic resonance imaging (MRI) showed a small remain in the pituitary parenchyma in the sella turcica floor, whose signal was typical of normal adenohypophysis, as well as sella turcica enlargement. This pituitary atrophy and a large sella turcica were defined as consistent with PA. Radiographic studies performed two years later showed evolution to an empty sella turcica with no other changes. The patient is currently asymptomatic and has no complications secondary to replacement therapy. In most cases of PA, the underlying pathological process is a pituitary tumor (known either before or after PA occurrence). Although PA is an uncommon condition, various studies have shown the existence of factors precipitating its development in up to 40% of patients^{2,3}: high blood pressure, head trauma, anticoagulant therapy, radiation therapy, a history of major surgery (specially heart surgery), or procedures such as dynamic pituitary function tests, general anesthesia, or coronary perfusion scintigraphy, in a recently reported case.⁴ The pathophysiology of PA has not been fully elucidated yet. For PA occurring on an adenoma, there are several theories, including rapid tumor growth exceeding blood supply and causing necrosis, or potential vasculopathies intrinsic to pituitary tumors (because these bleed up to 5.4 times more than other central nervous system tumors).⁵

In the reported case, tooth extraction and subsequent bleeding were considered to be the most likely causes of pituitary infarction and the subsequent development of hypopituitarism. The patient's history revealed no causes of hypopituitarism such as neurosurgical procedures or radiation therapy, a history of head trauma or compression symptoms, of hormone hypo- or hyperfunction, suggesting pituitary or suprasellar tumor. There was also no history or clinical signs consistent with systemic or infectious infiltrative disease. We think that this was a particularly uncommon case because PA did not occur in the setting of a pituitary adenoma. In addition, the patient had none of the precipitating factors associated with PA. Etiology is however the most unique characteristic. After a systematic review, we found no case reported in the scientific literature where the only triggering factor of PA was bleeding after tooth extraction in a healthy patient with no concomitant diseases or treatment, or during the performance of certain procedures. The pathogenesis of PA in our patient may

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be similar to that attributed to Sheehan's syndrome: sharp and sudden blood pressure changes due to profuse bleeding may cause arterial spasm in small vessels and trigger PA. Unlike as occurs in Sheehan's syndrome, where lactotroph cells experience physiological hyperplasia, stimulated by estrogens secreted by the placenta (which leads to 30–100% increases in pituitary weight),⁶ our patient showed no pituitary gland changes. As discussed above, MRI of the patient showed no tumor remains or other space-occupying lesions, except for bleeding consistent with PA. PA, and especially its subclinical presentation, continues to be a diagnostic challenge, not only because of the low frequency of the disease, but also because of the variability in clinical manifestations. Indeed, the condition only starts with its typical symptoms (headache, nausea and vomiting, ocular palsy, or visual field or acuity defects) in 3% of patients with pituitary adenomas. Because of the unusual circumstances of our patient, diagnosis was not made until four years after the event that triggered PA. It should be stressed that pituitary failure, specifically ACTH deficiency, has a strong impact on the morbidity and mortality of patients with PA if early diagnosis is not made and adequate replacement therapy is not started.

References

1. Sibal L, Ball SG, Connolly V, James RA, Kane P, Kelly WF, et al. Pituitary apoplexy: a review of clinical presentation, management and outcome in 45 cases. *Pituitary*. 2004;7: 157–63.
2. Rajasekaran S, Vanderpump M, Baldeweg S, Drake W, Reddy N, Lanyon M, et al. UK guidelines for the management of pituitary apoplexy. *Clin Endocrinol (Oxf)*. 2011;74:9–20.
3. Vicente A, Lecumberri B, Gálvez MA. Guía de práctica clínica para el diagnóstico y tratamiento de la apoplejía hipofisaria. *Endocrinol Nutr*. 2013;60, 582.e1–12.
4. Ferreira-González L, Trigás-Ferrín M, Rubin EP, Sesma P. Pituitary apoplexy after myocardial perfusion scintigraphy. *Endocrinol Nutr*. 2012;59:606–8 [article in English, Spanish].
5. Randeve HS, Schoebel J, Byrne J, Esiri M, Adams CB, Wass JA. Classical pituitary apoplexy: clinical features, management and outcome. *Clin Endocrinol (Oxf)*. 1999;51:181–8.
6. Kelestimur F. Sheehan's syndrome. *Pituitary*. 2003;6:181–8.

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