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SCIENTIFIC LETTERS

An unexpected sella turcica tumor☆

Tumor inesperado en la silla turca

Visual disturbances and hypopituitarism are not exclusive symptoms of pituitary macroadenomas. Other tumors clinically behaving as macroadenomas may occur in the sellar area. The most common of such tumors include craniopharyngioma, Rathke pouch cyst, and meningioma.[1] The case of a female patient in whom a pituitary macroadenoma was initially suspected but who was finally diagnosed with another sellar tumor is reported below.

This was a 49-year-old female patient born in the Philippines with a history of hypertriglyceridemia treated with gemfibrozil 900 mg/day and high blood pressure treated with indapamide 2.5 mg/day. She was referred to our clinic by her primary care physician for an impaired thyroid function in two consecutive laboratory tests showing decrease free tetraiodothyronine (T4) levels and normal thyroid-stimulating hormone (TSH) levels (first laboratory tests: TSH 3.26 μIU/mL and free T4 0.50 ng/mL; second laboratory tests: TSH 2.74 μIU/mL and free T4 0.60 ng/mL; normal ranges were 0.465–4.68 μIU/mL for TSH and 0.78–2.19 ng/dL for free T4 respectively). Patient reported a self-limited episode of galactorrhea and mastodynia 8 years before. She had been examined in her country, but had no medical reports, and stated that no cause had been found. In the past 2 years, the patient had experienced very severe asthenia, facial edema, and amenorrhea. She was also being monitored at the ophthalmology department for decreased visual acuity. A computed campimetry performed revealed bitermporal hemianopsia, and the ophthalmologist had therefore requested magnetic resonance imaging (MRI) which had not been performed yet. Facial edema, particularly of the eyelids, was conspicuous. The thyroid gland could be palpated, but no nodules were felt. There were no other remarkable findings in the physical examination or galactorrhea. A pituitary macroadenoma was suspected, and repeat measurements of pituitary basal hormones and assessment of MRI were therefore decided. MRI

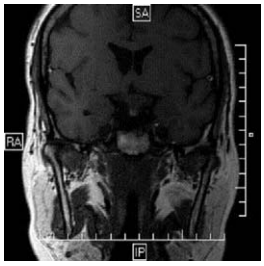

Figure 1 Thick-section initial magnetic resonance imaging of the skull suggesting pituitary macroadenoma.

of the brain (Fig. 1) showed a lesion with its center in the pituitary gland with a significant suprasellar component that appeared homogeneously hypointense in T1 and T2 with no presence of calcium foci, necrosis, or hemorrhage. The lesion was 25 × 24 × 26 mm in diameter, showed no signs of invasion of cerebral parenchyma, and did not invade sphenoidal sinus. The lesion extended laterally encompassing completely the left internal carotid and supraclinoid arteries, and partially the right internal carotid artery, which had a normal size with a signal void, which suggested patency. The chiasm was not adequately visualized, which was considered to be due to chiasm invasion. However, sections were too thick for adequate definition of this structure. Laboratory tests showed, in addition to pituitary hypothyroidism, hypopituitarism of the somatotropic and gonadotropic axes, as well as moderate hyperprolactinemia: follicle-stimulating hormone (FSH) 3.62 mIU/mL (normal range (NR), 21.5–131), luteinizing hormone (LH) 0.44 mIU/mL (NR: 13.1–86.5), 17-beta-estradiol 9.48 pg/mL (NR: 5.3–38.4), prolactin: 53.90 ng/mL and 59.50 ng/mL in the first and second samples respectively (NR: 3–25), growth hormone (GH) <0.05 ng/mL (NR <8.6), adrenocorticotropic hormone (ACTH) 49.90 pg/mL (NR: 4.7–48.8), cortisol 18.80 mg/dL (NR: 5–25) and insulin-like growth factor 1 (IGF-1) 81.45 ng/mL (NR: 90–360). Treatment was started with levothyroxine 25 μg daily for 2 weeks and 50 μg thereafter. Although the tumor was initially considered to be a macroadenoma, suprasellar extension suggested that it could be a meningioma, and repeat MRI was performed, but this time focused on the pituitary gland and using sections less than 3-mm thick. MRI (Fig. 2) revealed persistence

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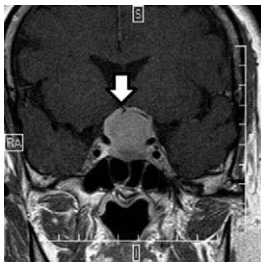

Figure 2 Thin-section magnetic resonance imaging centered on the pituitary gland showing the dural tail characteristic of meningioma.

of a lesion centered in the pituitary gland with a significant suprasellar component, homogeneously hypointense in T1 and T2 with no apparent calcium foci, necrosis, or hemorrhage. The lesion extended laterally encompassing completely the left internal carotid and supraclinoid arteries, and partially the right internal carotid artery. Optic chiasm was cranially reflected, with a slight deformity in third ventricle floor. Anteriorly, lesion extended toward the sphenoidal plane at an obtuse angle. After contrast administration, a small dural tail was associated (arrow in Fig. 2). Posteriorly, the lesion reached the prepontine cistern with no brain stem deformity. T1 hypersignal of the neurohypophysis was seen, adjacent to which there was a slightly more intense contrast enhancement that appeared to correspond to the compressed adenohypophysis. The pituitary stalk was not clearly identified. Signal homogeneity and extension to the sphenoidal plane and presence of dural tail suggested meningioma of sellar diaphragm as first diagnostic possibility.

Patient underwent surgery consisting of bifrontal craniotomy. Wide tumor excision was performed, completely decompressing the optic chiasm and leaving residual tumor fully encompassing the anterior communicating artery and both anterior cerebral arteries. Histological report confirmed suspicion of meningothelial meningioma, describing psammoma bodies, a fibrohyaline reaction, and absence of architectural changes, cytological atypia, and mitotic activity.

Preoperative temporal hemianopsia of the right eye persisted after surgery, and patient additionally experienced amaurosis in her left eye, although in the last visits she was already able to see light through this eye. She had partial central diabetes insipidus, which was controlled with oral desmopressin 0.1 mg and persisted in subsequent visits. Control laboratory tests showed, in addition to the abovementioned deficits and hyperprolactinemia, decreased basal cortisol levels and inappropriately normal ACTH levels: FSH 2.12 mIU/mL, LH <0.216 mIU/mL, prolactin 69.60 ng/mL, ACTH 13.82 pg/mL and basal cortisol 0.29 μg/dL, TSH <0.015 μIU/mL, and free T4 1.18 ng/dL.

Meningiomas are the most common benign tumors in central nervous system. They arise from arachnoid meningeal cells and may occur in any location of the CNS. Sellar diaphragm is a fold that is perforated for passage of sella turcica. This fold extends over the sellar tubercle to the top of dorsum sellae and clinoid process. Meningiomas arising from this dura mater area grow toward the sella turcica, occupying it and mimicking a pituitary adenoma.[2] The concept of meningioma of sellar diaphragm is relatively recent. In their 1938 monograph «Meningiomas», Cushing and Eisenhardt reported meningiomas with these characteristics, but classified them as suprasellar meningiomas. In 1954, Bush and Mahneke reported 25 patients who had undergone surgery for meningioma of sellar diaphragm.[3] Finally, in 1995 Kinjo et al.[4] proposed that these tumors are classified as type A when they arise from the upper leaf of sellar diaphragm and grow centrally to the sella turcica; as type B when they arise from the same but grow dorsally; and as type C when they are derived from the lower leaf and grow toward the sella turcica. The latter are the ones that clinically behave as pituitary macroadenomas.

Differential diagnosis between pituitary macroadenoma and meningioma of sellar diaphragm is very important because the surgical approach is completely different. Current advances in MRI facilitate distinction, but careful inspection of images is required in cases such as the one reported. Cappabianca et al.[5] reviewed MRI images from patients who had been diagnosed with non-functioning macroadenoma when they actually had meningioma of sellar diaphragm. Pituitary gland was always clearly visualized in meningiomas, which showed a bright, homogeneous enhancement after contrast administration, while a poor, heterogeneous enhancement was seen in macroadenomas. The center of the lesion was suprasellar in meningiomas and sellar in macroadenomas. Macroadenomas enlarged the sella turcica. However, meningiomas moderately enlarged sella turcica proportionately to their size. Finally, sellar diaphragm and pituitary stalk were identified in most patients with meningioma.

Meningiomas of sellar diaphragm should be included in differential diagnosis in patients with hypopituitarism and visual disturbances. Their preoperative identification is particularly important because surgical approach will vary.

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Postpartum hyponatremic coma[☆]

Coma hiponatrémico posparto

A 33-year-old primigravida was admitted to our center for hyponatremic coma 7 days after delivery of a healthy infant. She had no remarkable personal or family history, and was admitted to a private hospital for delivery after an uneventful full term pregnancy. High blood pressure was found upon admission, and a cesarean section was therefore decided, which was complicated by heavy bleeding due to a right uterine ligament tear. Subsequent laboratory tests showed AST levels of 100 U/L (5–40), anemia (hemoglobin 6.5 g/dL [12–16]), thrombocytopenia (21×10 E3/μL, 150–450), and proteinuria in the nephrotic syndrome range. Four RBC units and seven platelet units were transfused, and urapidil, labetalol 200 mg/6 h, and methyldopa 500 mg/6 h were infused due to sustained HBP. A single dose of cabergoline 1 mg was administered to inhibit lactation on the patient decision.

Seven days after birth, patient reported severe asthenia and headache. Physical examination showed no changes and BP values of 150–155/95–100 mmHg. A complete blood count revealed persistent anemia. A few hours later the patient experienced a tonic–clonic seizure followed by coma and was intubated. She remained afebrile and hemodynamically stable, with a BP of 135/80 mmHg and a HR of 67 bpm. Laboratory tests revealed worsening of anemia, blood glucose of 73 mg/dL, CPK of 1710 U/L related to the seizure, and Na of 103 mmol/L. Patient was transferred to the ICU of our center because of the need for imaging tests.

Upon arrival to the ICU, sodium levels of 104 mmol/L (135–145) were found, as well as glucose 78 mg/dL (60–100), urea 19 mg/dL (15–50), creatinine 0.57 mg/dL (0.5–1.2), and serum osmolality of 224 mOsm/kg. Urinary analysis showed Na 41 mmol/L and osmolality of 384 mOsm/kg. There was persistent normocytic and normochromic anemia with hemoglobin 8.2 g/dL, and slightly elevated transaminase levels. Chest X-rays and a CT scan of the head showed no changes, and diffuse brain involvement was found in an electroencephalogram. Isotonic saline, 2000 mL over 5 h, was started at the ICU with no improvement. Urine output during the first admission day ranged from 15 to 50 mL/h. Hormone tests requested provided the following values: cortisol 1.6 ng/dL (5–25), which increased to 19.1 ng/dL (30′) and 18.7 ng/dL (60′) after short stimulation with intravenous ACTH 250 mcg, TSH 0.56 mU/L (0.34–5.6) and free T4 0.57 ng/dL (0.58–1.64). Adrenal insufficiency was suspected, and replacement was therefore started with hydrocortisone 100 mg every 6 h. Urine output increased 2 h after each dose (145 mL after the first dose, 470 mL after the second, and 880 mL after the third), but urine output again decreased a few hours after each dose, and a continuous hydrocortisone infusion was therefore started.

On the second day, patient had normal Na at 6:00 h. At 9:00 h, repeat hormone tests showed a free T3 value of 1.8 pg/mL (2.5–3.9) and still decreasing TSH and free T4 levels. At 12:00 h, Na level increased to 119 mmol/L (11 mmol/L in 6 h) and urine output to 300–400 mL/h, and intravenous desmopressin 1 mcg was therefore administered (Fig. 1).

Laboratory test results on the third day included: hemoglobin 8.3 g/dL, glucose 112 mg/dL, creatinine 0.47 mg/dL, uric acid 0.8 mg/dL (2.5–6), Na 120 mmol/L, K 4.2 mmol/L, LDH 869 U/L, and normalization of transaminase levels. Hormone tests showed the following: prolactin 2 ng/mL, undetectable LH and FSH, cortisol 41.4 ng/dL, TSH 0.34 mU/L, free T3 1.87 pg/mL, and free T4 0.52 ng/dL. During the third and fourth days, sodium levels gradually increased to 138 mmol/L, with 10 mmol/L changes in less than 24 h and urine output values higher than 300 mL/h. Desmopressin 1 mcg was therefore administered again.

Deintubation was decided based on recovery of sodium levels, and patient remained conscious and oriented, with no focal neurological signs or new seizures. MRI showed a pituitary gland of normal shape and size, with peripheral enhancement and no central uptake with gadolinium contrast, consistent with non-hemorrhagic subacute adenohypophyseal ischemia. Sodium levels remained stable, and a gradual improvement occurred in thyroid hormone levels (free T4 0.62 ng/dL on the sixth day).

Patient was transferred to the endocrinology ward. Improvement in BP levels and disappearance of proteinuria allowed for tapering of antihypertensive drugs. Based on improved hormone control, decreased ischemia in the control MRI, and negative immune tests, corticosteroid tapering was decided. On hospitalization day 27, hormone tests were again performed before discharge, showing basal cortisol of 9 ng/dL, increasing to 14.8 ng/dL (30′) and 5.5 ng/dL (60′) after short stimulation with intravenous ACTH 250 mcg, and improvement in all other parameters. It was therefore decided to prescribe corticosteroid therapy under stress conditions only (Fig. 2).

After discharge, patient did not still require background treatment. Menses returned at 5 months of discharge, and

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