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Original article

Stroke disclosing primary aldosteronism: Report on three cases and review of the literature

Accident vasculaire cérébral révélant un hyperaldostéronisme primaire : à propos de trois cas et revue de la littérature

Amina Nasri [a,*], Malek Mansour [a], Zeineb Brahem [a], Amel Kacem [b], Ahmed Abou Hassan [a], Hager Derbali [a], Meriem Messelmani [a], Jamel Zaouali [a], Ridha Mrissa [a]

[a] *Department of Neurology, Military Hospital, 1008, Montfleury, 1089 Tunis, Tunisia*
[b] *Department of Medicine, Regional Hospital of Jendouba, Tunisia*

Abstract

Objectives. – There is a growing evidence of increased risk of cerebrovascular events in primary aldosteronism (PA). Nevertheless, acute neurologic ailment as presenting feature of PA is uncommon. Our aim is to highlight the diagnosis challenges in stroke unmasking PA and to discuss the underlying physiopathology and management dilemmas. *Materials and methods.* – We hereby describe three consecutive rare cases of stroke revealing PA. All patients had brain imaging and thorough biological and morphological assessment to rule out other etiologies of stroke. The diagnosis of primary aldosteronism was established according to the Endocrine Society Clinical Practice Guideline, with a review of the literature on the spectrum of neurologic manifestations in PA. *Results.* – We report on three cases, two women and a man, presenting with ischemic or hemorrhagic stroke, of early onset in two of them. All of the reported patients had hypertension and hypokaliemia. This association prompted the assessment of renin angiotensin aldosterone system (RAAS) disclosing PA, which was due to bilateral adenomas in the first one or bilateral adrenal hyperplasia in the two others. All patients refused the surgical option and received spironolactone with recurrence of stroke in one of them due to treatment incompliance. *Conclusion.* – Although cerebrovascular events are quite common in PA, their occurrence as initial feature can be misleading. The association of hypokaliemia and refractory hypertension in ischemic or hemorrhagic strokes should prompt an assessment of the RAAS to rule out PA and initiate adequate management as soon as possible in order to avoid further complications.

Keywords: Aldosteronism; Stroke; Ischemic; Hemorrhage

Résumé

But/objectif. – Le risque accru d'évènements cérébro-vasculaires est de plus en plus reconnu dans l'hyperaldostéronisme primaire (HAP). Cependant, la survenue inaugurale de manifestations neurologiques aiguës demeure inhabituelle dans l'HAP. Notre objectif était de mettre en évidence les difficultés diagnostiques des accidents vasculaires cérébraux (AVC) révélant un HAP et d'en discuter les mécanismes physiopathologiques et la prise en charge. *Matériels et méthodes.* – Nous décrivons trois cas d'AVC révélant un HAP. Tous les patients avaient eu une imagerie cérébrale et un bilan biologique et morphologique exhaustif afin d'éliminer toute autre cause d'AVC. Le diagnostic d'HAP a été établi selon les recommandations de l'Endocrine Society Clinical Practice Guideline, avec revue de la littérature du spectre de troubles neurologiques dans l'HAP. *Résultats.* – Nous rapportons trois cas, deux femmes et un homme, se présentant avec un tableau d'AVC ischémique ou hémorragique inaugural, à début précoce chez deux d'entre eux. Tous avaient une hypertension artérielle (HTA) et une hypokaliémie. Devant cette association, l'étude du système rénine-angiotensine-aldostérone (SRAA) avait révélé un HAP, dû à des adénomes surrénaliens bilatéraux dans le premier cas ou à une

* Corresponding author. Hôpital Militaire Principal d'Instruction de Tunis, 1008, Montfleury, C.P.: 1089, Tunis, Tunisia.
E-mail addresses: dr.nasri.amina@gmail.com, dna3006@gmail.com (A. Nasri).

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hyperplasie surrénalienne bilatérale chez les deux autres. Tous les patients ont refusé la chirurgie et ont donc reçu la spironolactone avec une récidive hémorragique chez l'un d'entre eux due à une mauvaise observance du traitement antihypertenseur. *Conclusion.* – Malgré la fréquence des évènements cérébro-vasculaires dans l'HAP, leur survenue comme manifestation inaugurale peut être déroutante. L'association d'une hypokaliémie et d'une HTA réfractaire dans les AVC ischémiques et hémorragiques doit faire pratiquer une étude du SRAA à la recherche d'un HAP et initier la prise en charge adéquate afin d'éliminer d'autres complications.

Mots clés : Hyperaldostéronisme ; Accident vasculaire cérébral ; Ischémie ; Hémorragie

1. Introduction

Primary aldosteronism (PA), first described by Conn in 1955 [1], is defined as autonomous overproduction of the mineralocorticoid hormone aldosterone, leading to resistant hypertension with or without hypokalemia [2,3]. PA can be due to aldosterone-producing adenomas, bilateral adrenal hyperplasia [3], or more rarely, to adrenocortical carcinoma. While the majority of cases of PA are sporadic, forms of familial hyperaldosteronism have been also described [4]. Currently, there is a growing evidence of increased risk of cardiovascular and cerebrovascular events in PA [5,6].

2. Methods

We hereby describe three consecutive rare cases of stroke revealing PA and discuss their underlying physiopathology, diagnosis challenges and management dilemmas with a review of the literature.

3. Results

3.1. Case 1

A 58-year-old man, with no peculiar family or personal history, presented with sudden onset weakness of the right limbs and slurred speech. On examination, he had drowsiness, Broca's aphasia and right hemiparesis with persisting high blood pressure. Brain CT scan showed left capsulolenticular hematoma (Fig. 1A). The transthoracic echocardiography (TTE), holter monitor (HM), carotid and vertebral Doppler ultrasound (CVDU) were all normal. Laboratory investigations revealed hypokalemia at 2.1 mmol/l (Normal: 3,5 to 5 mmol/l). In search of the cause of hypokalemia, we performed a simultaneous measurement of plasma aldosterone concentration (PAC) and direct renin (DR), in the morning, more than 2 hours after awakening, after 5 to 15 min of seated posture, and discontinuation of interfering medication. We found an increased PAC at 758 pmol/l (Normal: 8–172 pmol/l) with a low DR at 1.5 mIU/l (Normal: 2.8–39,9 mIU/l) and an elevated aldosterone-renin ratio (ARR) at 152 (Normal < 64). These findings were confirmed by a second assessment. An associated corticism was ruled out with normal 24 hour urine free cortisol level and midnight serum cortisol level. An abdominal CT-scan with CT angiography of the renal arteries showed no renal artery stenosis, but disclosed bilateral benign adenomas (Fig. 1B). The diagnosis of hemorrhagic stroke due to hypertension revealing primary aldosteronism, probably due to benign adrenal adenomas was established according to the Endocrine Society Clinical Practice Guideline (Funder et al., 2016). The patient first received intravenous nicardipine with electronic syringe driver then an association of oral spironolactone (200 mg/day) and nicardipine (150 mg/day). On follow-up, he had a normalization of his blood pressure and an improvement of his hemiparesis.

3.2. Case 2

A 47 year-old woman had family history of stroke and personal history of diabetes. At the age of 39, she had acute onset right hemiparesis and dysarthria. Brain CT showed ischemic stroke of the left posterior limb of the internal capsule. In addition, she had persistent severe high blood pressure. On the preliminary laboratory tests, she had hypokaliemia at 2 mmol/l. The HM, CVDU and TTE showed no abnormalities except for left ventricular hypertrophy in the latter. Immunologic and thrombophilia testing were normal. PAC was increased at 740 pmol/l with low DR at 0.17 mIU/l and increased ARR at 340 (Normal < 64). An abdominal CT-scan with CT angiography of the renal arteries showed bilateral adrenal hyperplasia without renal artery stenosis (Fig. 1D). She received spironolactone (200 mg/day), moxonidine (0.4 mg/day) and aspirin (250 mg/day) with an improvement of her motor deficit and her blood pressure. Seven years later, she had a second episode of left hemiparesis. The anamnesis disclosed incompliance to antihypertensive drugs. Brain CT-scan showed a right capsulolenticular hematoma (Fig. 1C). Antiplatelets were ceased and antihypertensive therapy reintroduced with normalization of her blood pressure.

3.3. Case 3

A 42 year-old woman had personal history of resistant non-explored hypertension since the age of 29. She was admitted for sudden onset left hemiparesis. Her brain CT scan and brain MRI showed ischemic stroke of the posterior limb of the right internal capsule and extensive leukoaraiosis (Fig. 1E and G). Brain magnetic resonance angiography (MRA) showed three intracranial aneurysms (Fig. 1H). The HM, CVDU and TTE showed no abnormalities except for left ventricular hypertrophy in the latter. Laboratory findings revealed hypokalemia (1.9 mmol/l).