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236 - ABERRANT 5TH4 RECEPTORS AS CAUSE OF SUBCLINICAL CUSHING&RSQUO;S SYNDROME DUE TO ACTH-INDEPENDENT BILATERAL MACRONODULAR ADRENAL HYPERPLASIA: CASE REPORT

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Resumen

Introduction: Bilateral macronodular adrenal hyperplasia (BMAH) is a rare cause (< 1%) of endogenous Cushing's syndrome (CS). Aberrant ectopic or eutopic receptors to GIP, catecholamines, luteinizing hormone or serotonin (5-HT_4) and many other receptors have been described as the cause of autonomous production of glucocorticoids by the adrenal cortex in this subtype of CS. Accordingly, in vivo protocols that study different responses to physiological and pharmacological tests have been developed for the screening of the involvement of such aberrant receptors in these particular CS cases.

Case report: A 66-year-old woman referred to our Department for the study of BMAH. After confirming the presence of ACTH-independent hypercortisolism by lack of suppression to dexamethasone and the presence of suggestive adrenal images, we performed a 4-day screening protocol to test functionally the presence of aberrant receptors. We found a positive response of cortisol to the stimulation with metoclopramide consisting of a 63% increase in serum cortisol concentrations, suggesting the presence of aberrant 5TH4 receptors in the surface of the membrane in the *zona fasciculata* of the adrenal cortex. Because the only commercially available drug with antagonist $5TH_4$ receptor properties is amitriptyline, we submitted the patient to metoclopramide stimulation test after a single 25 mg dose of oral amitriptyline. The abnormal serum cortisol response and the physiological aldosterone increase were partially blunted. Hence, we started a therapeutic trial with amitriptyline 75 mg once a day with no apparent immediate side effects.

Discussion: Screening for aberrant receptors in patients with BMAH and mild CS is mandatory as it may result into a chance to pharmacologic therapy as an alternative to adrenal surgery.