Introduction

We presented three cases of Cushing’s syndrome (CS) with bilateral adrenal tumors. When bilateral adrenal tumors are encountered, the differential diagnosis is difficult, especially in the functioning bilateral adrenocortical adenoma. Adrenal scintigraphy, which has become a standard technique to determine the laterality of excessive hormone secretion, showed bilateral adrenal activity in all cases. However, adrenal venous sampling (AVS) distinguished three unique hormonexcess patterns. Based on these findings, we could select adequate treatment for each case. Thus, AVS is necessary to obtain a definitive diagnosis and optimal therapy in CS with bilateral adrenal tumors.

Case A

Diagnosis

CS due to bilateral adrenal tumors

Treatment

To preserve adrenal function, the left adrenal gland was totally resected, whereas the right adrenal gland was partially resected laparoscopically.

Case B

Diagnosis

CS caused by the right adrenal tumor and PA caused by both adrenal glands

Treatment

We performed a laparoscopic right total adrenalectomy for the treatment of CS and offered medical therapy for the treatment of primary aldosteronism (PA).

Case C

Diagnosis

PA caused by both adrenal glands and subclinical CS caused by both adrenal tumors

Treatment

This patient has no risk factors for cardiovascular disease such as hypertension, obesity, diabetes mellitus or dyslipidemia. Thus, we are observing her without medical therapy.

Discussion

It is well known that AVS provides important information concerning the laterality of excessive aldosterone secretion [1]. However, the use of AVS in patients with CS due to bilateral adrenal adenomas has rarely been reported [3-8]. Adrenal scintigraphy showed bilateral adrenal activity in all cases. However, AVS demonstrated three different hormone-excess patterns. Based on these findings, we selected a different treatment approach for each case. Thus, AVS is necessary to obtain a definitive diagnosis and optimal therapy in CS with bilateral adrenal tumors.

Discussion 2

There is no consensus regarding the optimal determination of the laterality of excessive hormone secretion. Omura et al. demonstrated the criterion of super-selective ACTH-stimulated AVS for the laterality of cortisol secretion in CS [9]. This report indicated that the cortisol levels were >300 mcg/l in both cortisol veins or the tributary veins from the adrenals, and the cortisol levels were <300 mcg/l in other tributary veins in the case of bilateral cortisol-secreting adenoma. This criterion may have applied to our cases and may therefore serve as a standard criterion for AVS in CS with bilateral adrenal tumors.

References


Concluding remarks

We presented three cases of CS with bilateral adrenal tumors. When bilateral adrenal tumors are encountered, the differential diagnosis is difficult, especially in the functioning bilateral adrenocortical adenoma. Adrenal scintigraphy, which has become a standard technique to determine the laterality of excessive hormone secretion, showed bilateral adrenal activity in all cases. However, AVS distinguished three unique hormone-excess patterns. Based on these findings, we could select adequate treatment for each case. Thus, AVS is necessary to obtain a definitive diagnosis and optimal therapy in CS with bilateral adrenal tumors.