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- Fuertes Zamorano N, De Miguel Novoa M, Molino González A, Díaz Pérez J, Martín Rojas-Marcos P, Montañez Zorrilla M. Disfunción tiroidea en los pacientes con carcinoma de células renales avanzado en tratamiento con sunitinib: un problema multifactorial. Endocrinol Nutr. 2010.
- 3. Wong E, Rosen LS, Mulay M, Vanvugt A, Dinolfo M, Tomoda C, et al. Sunitinib induces hypothyroidism in advanced cancer patients and may inhibit thyroid peroxidase activity. Thyroid. 2007;17:351-5.
- 4. Mannavola D, Coco P, Vannucchi G, Bertuelli R, Carletto M, Casali PG, et al. A novel tyrosine kinase inhibitor, sunitinib, induces transient hypothyroidism by blocking iodine uptake. J Clin Endocrinol Metab. 2007;92:3531-4.
- Tamaskar I, Bukowski R, Elson P, Ioachimescu AG, Wood L, Dreicer R, et al. Thyroid function test abnormalities in patients with metastatic renal cell carcinoma treated with sorafenib. Ann Oncol. 2008;19:265-8.
- Torino F, Corsello SM, Longo R, Barnabei A, Gasparini G. Hypothyroidism related to tyrosine kinase inhibitors: an emerging toxic effect of targeted therapy. Nat Rev Clin Oncol. 2009;6:219-28.
- Wolter P, Stefan C, Decallonne B, Dumez H, Bex M, Carmeliet P, et al. The clinical implications of sunitinib-induced hypothyroidism: a prospective evaluation. Br J Cancer. 2008;99: 448-54.

- Garfield D, Hercbergs A, Davis P. Unanswered questions regarding the management of sunitinib-induced hypothyroidism. Nat Clin Pract Oncol. 2007:12:674-5.
- Garfield DH, Hercbergs A, Davis PJ. Hypothyroidism in patients with metastatic renal cell carcinoma treated with sunitinib. J Natl Cancer Inst. 2007;99:975-6.
- Porlan E, Vidaurre OG, Rodríguez-Peña A Thyroid hormone receptor b (TRb1) impairs cell proliferation by the transcriptional inhibition of cyclins D1, E and A2. Oncogene. 2008;27:2795-8.

Gilberto Pérez López^{a,*}, Marta Carrasco De La Fuente^b, Miriam Menacho Román^c, Olga González Albarrán^a, Marta Cano Megías^a

^aServicio de Endocrinología y Nutrición, Hospital Universitario Ramón y Cajal, Madrid, Spain ^bServicio de Endocrinología y Nutrición, Hospital La Moraleja, Madrid, Spain ^cServicio de Bioquímica Clínica, Hospital Universitario Ramón y Cajal, Madrid, Spain

*Corresponding author.

E-mail address: beto_med@hotmail.com (G. Pérez López).

Primary adrenalin sufficiency due to bilateral adrenal lymphoma

Insuficiencia suprarrenal primaria por linfoma adrenal bilateral

To the Editor:

A patient with adrenal insufficiency secondary to a bilateral primary adrenal lymphoma (PAL) is reported. A brief review of the literature on the most relevant clinical and pathological characteristics of this condition is also provided.

An 80-year-old female patient with a history of high blood pressure complained of constitutional symptoms, nausea, and vomiting during the previous four months. Supplementary tests performed revealed persistently elevated serum LDH levels (2,000 IU/L) and normal electrolyte levels. A CT scan of chest and abdomen (Fig. 1) showed masses in both adrenal glands. A study of the adrenal axis showed ACTH levels ranging from between 300 and 500 pg/mL (normal range, 10-50 pg/mL) and basal cortisol levels ranging from between 10 and 14 µg/dL (normal range, 3-18 µg/dL). Following glucocorticoid replacement therapy, laparotomy was performed, at which a biopsy could only be performed because both lesions were surgically non-resectable. A pathological study reported a large diffuse B-cell non-Hodgkin lymphoma, CD20 positive and Ki-67 positive in approximately 90% and P53 positive in approximately 80%. No bone marrow infiltration was found. Chemotherapy was started and well tolerated, and, at the time of writing, response to it is pending evaluation.

Primary lymphoma of the adrenal gland accounts for approximately 1% of extranodal lymphomas, and there are less

than 100 tumors of this type reported in the literature¹. It is more common in males than in females (2/1), and mean age at tumor occurrence is approximately 68 years². Bilateral adrenal involvement is found in 65% of cases³. Ninety percent of PALs are large B-cell non-Hodgkin lymphomas⁴.

The most common initial symptoms include abdominal pain, lumbar pain, fever, and weight loss. In patients with bilateral involvement, some degree of adrenal insufficiency may be found in up to 60% of cases, but insufficiency is usually subclinical in most of them.

Despite its low incidence, PAL should be included in differential diagnosis of an adrenal gland together with other malignant tumors such as carcinoma of the breast and lung, gastrointestinal tract tumors, and malignant melanoma, in which the incidence of adrenal metastases, often bilateral, is high. Differential diagnosis should also include other con-



Figure 1 CT of the chest and abdomen.

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ditions such as primary adrenal carcinoma, pheochromocytoma, and some infectious conditions⁵.

PAL is the disease with the poorest prognosis among extranodal B-cell lymphomas. Ninety percent of reported patients die within one year of diagnosis despite treatment⁶. The chemotherapy schemes routinely used with curative intent for the treatment of high-grade B lymphomas provide very poor results in PAL. Radiotherapy may improve local control of the disease, but its impact on the survival of these patients is unknown⁷. Finally, glucocorticoid replacement therapy should be part of the treatment when some degree of adrenal insufficiency exists.

References

- 1. Freeman C, Berg JW, Cutler SJ. Ocurrence and prognosis of extranodal lymphomas. Cancer. 1972;29:252-60.
- Wang J, Sun N, Renslo R, Chiang C, Tabarra HJ, Barajas L, et al. Clinically silent primary adrenal lymphoma: A case Report and review of the literature. Am J Hematol. 1998;58:130-6.
- Bauduer F, Delmer A, Tourneau AL, Guettier C, Alexandre JH, Zittoun R, et al. Primary adrenal lymphoma. Acta Haematol. 1992;88:213-5.

- Pimentel M, Jonson JB, Allan DR, Greenberg H, Bernstein CN. Primary adrenal lymphoma associated with adrenal insufficiency: A distinct clinical entity. Leuk Lymphoma. 1997;24:363-7.
- Singh D, Kumar L, Sharma A, Vijayaraghavan M, Thulkar S, Tandon N. Adrenal involvement in non-Hodgkin's lymphoma: Four cases and review of literature. Leuk Lymphoma. 2004;45:789-94.
- May F, Bachor R, Hack M, Gottfried HW, Hautmann RE. Primary adrenal non-Hodgkin's lymphoma: long-term survival. J Urol. 1998;160:487.
- Sasagawa I, Sadamori N, Itoyama T, Tsukasaki K, Nakamura H, Tomonaga M, et al. Primary adrenal lymphoma with chromosomal abnormalities. Acta Haematol. 1995;94:156-62.

Ignacio Rasero Hernández, Rafael Hernández Lavado*, Pilar Rodríguez Ortega, Carlos Guzmán Carmona, José Díaz Pérez de Madrid

Sección de Endocrinología y Nutrición, Hospital Infanta Cristina, Badajoz, Spain

*Corresponding author.

E-mail address: hernandezlavado@eresmas.com
(R. Hernández Lavado).