

reduce invasive monitoring, she wished to have LBA and underwent an uneventful procedure. She was discharged on fludrocortisone and hydrocortisone along with lanreotide for primary disease management. Metyrapone and spironolactone were stopped. At initial outpatient follow up, she had normal blood pressure and electrolytes. By her report, her quality of life had improved tremendously, given the reduced need for multiple clinic visits and blood investigations. Unfortunately, her primary disease progressed with gradual deterioration of physical function. She died six months after LBA but, as per her desire, spent her final months with loved ones. **Clinical Lesson:** In patients with EAS who have unresectable primary disease but otherwise good performance status, LBA is a viable alternative to medical management and should be discussed with patients early after diagnosis. References 1. Alberda et al., Endoscopic bilateral adrenalectomy in patients with ectopic Cushing's syndrome. *Surgical Endoscopy* 2012; 26:1140-1145 2. Reincke et al., A critical reappraisal of bilateral adrenalectomy for ACTH-dependent Cushing's syndrome. *Eur Journal of Endocrinology* 2015, 173M23-M32

Tumor Biology

TUMOR BIOLOGY CASE REPORTS

Late Onset of Abiraterone Severe Hypokalemia Due to Unsuspected ACTH-Secreting NEC

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Clinical Case: 68-year-old male, Active smoker (30-40 packs/year), and moderate alcohol drinker. An increase in urinary frequency, nocturia, and a PSA of 116 µg/L led to the diagnosis of disseminated castration-sensitive prostate cancer (CSPC), advanced Gleason score (3 + 4 pT2), with bone and retroperitoneal metastases (CT). By 02/2018, he started androgen deprivation therapy (ADT) with Abiraterone (AB) 1000 mg, prednisone, and LHRH agonist. 15 months later, PSA levels decreased to 0.75 ng/ml without side effects and normal K⁺(3.7 mEq/L), when a CT scan revealed both liver and bone metastasis. During the progression study, two months later, the patient was admitted to the hospital for severe hypoK⁺ (1.7 mEq / L), normal renal function and metabolic alkalosis. Although abiraterone was discontinued, up to 460 mEq iv per day of K⁺ and spironolactone were required to maintain serum K⁺ above 2.5. 3 days later, the hormonal study revealed TSH 1.13 mU /L, ACTH 162 pmol/L, cortisol 258 nmol/L, aldosterone 105 pmol/L, renin (protein) 0.7 µU/mL and deoxycorticosterone 507 pmol/L. 11 days later, plasma cortisol was 967 nmol/L, ACTH 132pmol/L, and cortisoluria 1456 nmol/d. The suppression test with 1 mg of DXM for cortisol was 1162,5 nmol/L. DHEAS was < 407 nmol/L. Liver biopsy showed a small cell neuroendocrine carcinoma (NEC), chromogranin (+), synaptophysin (+), CD56 (+), TTF-1 (+), PSA (-), Ki-67 90% and the granular cytoplasmic ACTH (+). The octreotide scan (+) revealed pathological uptake in L4, multiple uptakes in the liver, and in the axial skeleton. Treatment with Carboplatin plus Paclitaxel for NEC was started, completing 3 weekly doses with good tolerance and clinical benefit, but with the persistence of severe

hypoK⁺ (2.5 mEq/L). Treatment with lanreotide 120 mg every 4 weeks was added, following a feeling of clinical improvement, the disappearance of edema, asthenia, and normalization of plasma K⁺ (3,4mEq/L). The patient died two months later from respiratory sepsis. **Discussion:** Several clinical trials have demonstrated that the combination of AB plus ADT prolongs overall survival in DCSPC. The CYP17 inhibition by AB increases ACTH leading to early secondary mineralocorticoid excess with hypokalemia and hypertension while the cortisol levels remain normal or low. Cortisol serum levels increased after AB was discontinued, whereas aldosterone serum levels remained low due to K⁺ regulatory feedback. The hormonal profile and the pathological and radiological studies revealed an ACTH-producing small cell NEC. In this patient, the previous treatment with BA has masked the clinical and hormonal profile of an ectopic Cushing syndrome. Therefore, Cyp17 inhibitors can mask adrenal or extra-adrenal processes characterized by alterations in steroid metabolism. This case has suggested a more thorough assessment of the adrenal hormonal profile including ACTH during BA treatment.

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Multifocal Multisystem Langerhans Cell Histiocytosis in an Adult Female Atypically Presenting With Multiple Abdominopelvic Abscesses

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Introduction: Langerhans Cell Histiocytosis (LCH) is a condition of malignant clonal proliferation of myeloid bone marrow cells that more commonly affects children than adults. Furthermore, its presentation remains variable ranging from single organ to multisystem involvement including bone, skin, lymph, liver, spleen, lung, and central nervous system. We report an unusual case of multisystem LCH in an adult female presenting with multiple soft tissue abscesses.

Case: We report a case of a 38-year-old female with a past medical history of polysubstance abuse, type 2 diabetes, polycystic ovarian syndrome, and isolated central diabetes insipidus who had multiple hospitalizations for recurrent soft tissue abscesses treated with incision and drainage and antibiotics. Imaging studies revealed multiple osteolytic lesions involving the bilateral iliac crests, acetabulum and femur, as well as an iliopsoas abscess. Given her prior history of isolated central diabetes insipidus, the possibility of LCH as the cause was entertained. Histological evaluation performed on an inguinal soft tissue sample stained positive for CD1a and S100, and a formal diagnosis of Langerhans Cell Histiocytosis (LCH) was made. The patient was treated with chemotherapy with good results.

Conclusion: This report presents a rare and unusual case of adult onset multisystem LCH involving bone, skin, lymph, and central nervous system presenting with recurrent large abdominopelvic abscesses. These abscesses may represent a rare and unrecognized form of soft tissue involvement of LCH.