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Sudden onset of sarcoidosis after successful surgical treatment of Cushing's syndrome

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Sarcoidosis is a multisystem granulomatous disease with putative autoimmune aetiology, affecting mainly the lungs, but the eyes and skin may also be affected. Cushing's syndrome is characterized by excessive glucocorticoid (GC) secretion, leading to immunosuppression. Herein, we report the sudden activation of sarcoidosis upon normalization of the cortisol level after successful treatment for Cushing's syndrome.

A 43-year-old male was admitted to the Department of Endocrinology and Metabolic Disease with a right adrenal tumour that was discovered accidentally during an examination of chest computed tomography (CT) on 6 May 2021. Apart from the right adrenal tumour, chest CT (Fig. 1) did not reveal any abnormalities in the lungs and mediastinum. The patient had a 12-month history of hypertension (170/100; 166/108 mmHg), weight gain (12 kg) and asthenia. On a physical examination, the patient was obese [112 kg, body mass index (BMI) — 34.57 kg/m^2] with predominantly central disposal of fat tissue and abdominal purple striae.

Laboratory evaluation revealed normal glucose levels, complete blood count, and normal liver and renal functions. Screening functional tests were performed to eliminate Conn's syndrome, Cushing's syndrome, and pheochromocytoma. Twenty-four-hour urinary free metanephrine and normetanephrine levels were both within normal limits. Plasma aldosterone/plasma renin activity ratio was < 20 with normal serum potassium levels. The patient had a high midnight serum cortisol level (5.8 ug/dL), and the cortisol level was not suppressed with 2-day 2 mg dexamethasone suppression test. Basal adrenocorticotropic hormone (ACTH) levels were < 5 pg/mL at 3 time points during the day, indicating an ACTH-independent cause that contributed to decreased concentration of dehydroepiandrosterone sulphate (DHEAS). The adrenal CT confirmed

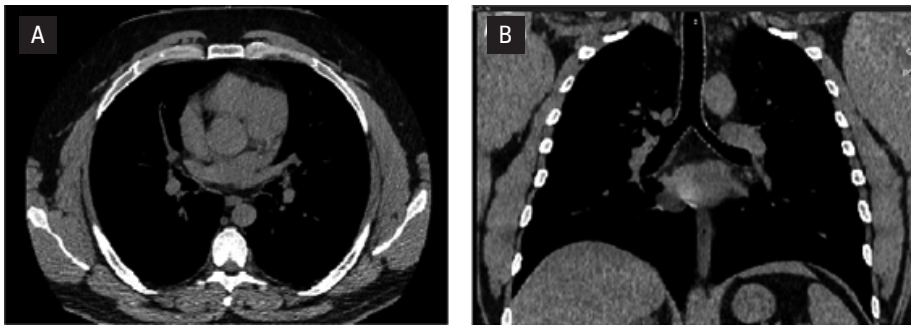

Figure 1. *Computed tomography (CT) scans showing normal mediastinal lymph nodes before laparoscopic adrenalectomy.* **A.** *Transverse plane;* **B.** *Coronal plane*

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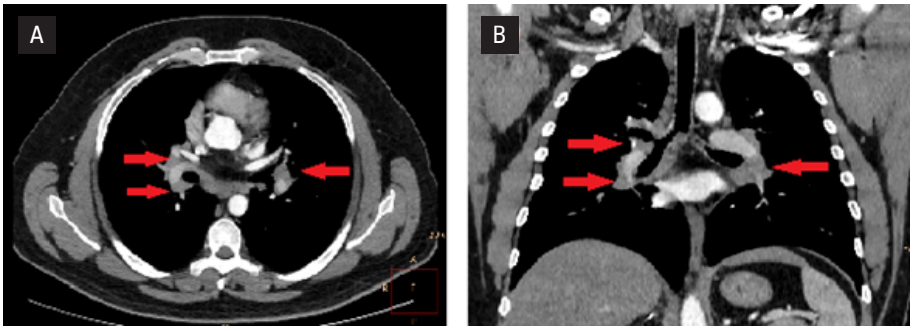

Figure 2. *Computed tomography (CT) scans showing enlarged mediastinal lymph nodes after laparoscopic adrenalectomy.* **A.** *Transverse plane;* **B.** *Coronal plane*

a presence of a right adrenal low-density mass, which was 37 × 31 × 31 mm in size and was indicative of an adenoma. On the basis of the above results, Cushing's syndrome was diagnosed. The patient underwent a right laparoscopic adrenalectomy in December 2021. The postoperative period was uneventful. He was discharged without temporary corticosteroids replacement. A pathomorphology examination revealed that the adrenal mass was a cortical adenoma, one point on the Weiss scale. After adrenalectomy, blood pressure normalized and the patient lost 10 kg. However, a few days after adrenalectomy, the patient developed generalized arthralgias and malaise. Symptoms were symmetric and oligoarticular, with increasing pain after rest. Therefore, the patient was readmitted to the Department of Endocrinology and Metabolic Disease on 30 March 2022, and hydrocortisone therapy 15 mg per day was introduced. Furthermore, a chest CT (Fig. 2) showed the multiple enlarged hilar and mediastinal lymph nodes. The video-assisted thoracoscopic surgery on the right side was performed on 28 May 2022, and a diagnosis of sarcoidosis was made. Ophthalmic and skin sarcoidosis was excluded. The patient felt well on hydrocortisone treatment, so the dose of the drug was not changed. The patient did not require any other causal treatment.

Oral GCs are the first-line treatment of sarcoidosis due to suppression of inflammatory response genes and inhibition of pro-inflammatory cytokine production. This makes GCs effective in controlling a wide range of autoimmune diseases that may also include sarcoidosis. In this case, the excessive elevation of GC levels in Cushing's syndrome apparently masked the sarcoidosis.

So far, there have been only a few cases of sarcoidosis after surgical treatment of Cushing's syndrome [1, 2] or Cushing's disease [3] reported in the literature. In previous studies, the mean time delay to onset of symptoms was several months; however, in our case the onset of the disease was after a few days. In our opinion, it was due to the lack of GC substitution after adrenalectomy. Unspecific symptoms made differential diagnosis with transient postoperative adrenal insufficiency very difficult. The case of our patient highlights the importance of strict follow-up care of patients after adrenalectomy. In our opinion, it is crucial to spread the knowledge amongst endocrinologists, resulting from our case, about the risk of development of some diseases, such as sarcoidosis, as a consequence of the normalization of the cortisol level.

Statements of ethics

The research was conducted ethically in accordance with the World Medical Association Declaration of Helsinki. Data were collected retrospectively. Written informed consent was obtained from the patient for the publication of this case report (including all laboratory data and images).

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Conflict of interests

The authors declare that they have no conflict of interests.

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