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Case report

A challenging case of laparoscopic synchronous bilateral adrenalectomy for Cushing's disease. Case report

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ABSTRACT

Introduction: Laparoscopic adrenalectomy is a treatment option in patients with Cushing's syndrome. Preoperative comorbities as well as surgical and anesthesiological difficulties can make the procedure challenging.

Presentation of the case: We present the case of a 53-year-old obese man diagnosed with Cushing's syndrome, also suffering from other endocrine pathologies, neurofibromatosis type 2, cardiomiopathy with severe hypertrophy and diastolic dysfunction, deep vein thrombosis (DVT) and obstructive sleep apnea syndrome (OSAS).

After multidisciplinary team discussion of the case, the patient underwent laparoscopic synchronous bilateral adrenalectomy. The laparoscopic approach was a part of a balanced enhanced recovery program which resulted in uneventful discharge in 4 days.

Conclusion: Laparoscopic synchronous bilateral adrenalectomy is feasible and effective and should be considered also in patients with wide preoperative comorbidities and challenging intraoperative management, as long as the patient is meticulously studied preoperatively. An approach including a multidisciplinary team discussion is recommended.

1. Introduction

Laparoscopic adrenalectomy is a standard of care in patient diagnosed with adrenal masses, except in case of involvement of surrounding tissue or adrenal and caval vein by a malignant adrenal lesion. Also, it is considered a treatment option in patients with Cushing's syndrome, specially in case of young age at diagnosis, due to difficulties in the management of medical therapy.

We present the case of a patient treated with laparoscopic synchronous bilateral adrenalectomy for Cushing's syndrome. The case is particularly interesting, due to its wide preoperative comorbidities and the challenging surgical and anesthesiological management.

We postulate that the ability to mitigate our patient's comorbidities and perioperative risk factors with a bilateral laparoscopic resection are as important as the more surgery-specific advantages.

This work has been written in accordance with SCARE criteria [1].

1.1. Case report

A 52-year-old Caucasian male with Cushing's syndrome was diagnosed with bilateral adrenocortical hyperplasia on CT-scan.

Medical history was consistent with type-2 neurofibromatosis. Consistently with his syndrome, he had been diagnosed with a pontocerebellar schwannoma treated with open and gamma-knife surgery and radiotherapy around 4 years prior to presentation; additionally, a lumbar spinal neurinoma had been diagnosed but managed conservatively. Toxic multinodular goitre was also present, for which medical management had proven difficult in the past. He had undergone multiple deep vein thromboses of the lower extremity, and a caval filter had been placed before brain surgery, which was partially occluded with chronic thrombus at the time of our intervention, despite chronic anticoagulation.

During follow-up and workup for possible spinal surgery, computed tomography (CT) had revealed bilateral adrenal hyperplasia without evidence of an ectopic ACTH-producing tumor (Figs. 1 and 2).

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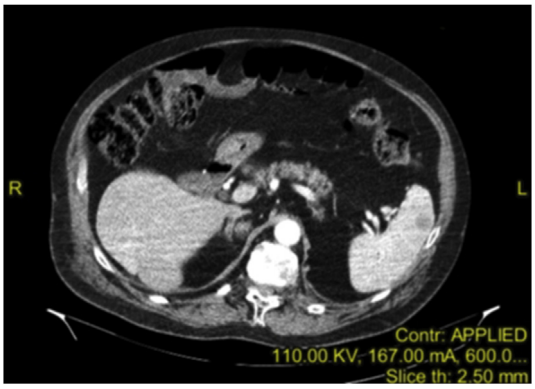

Fig. 1. Right adrenal on CT-scan.

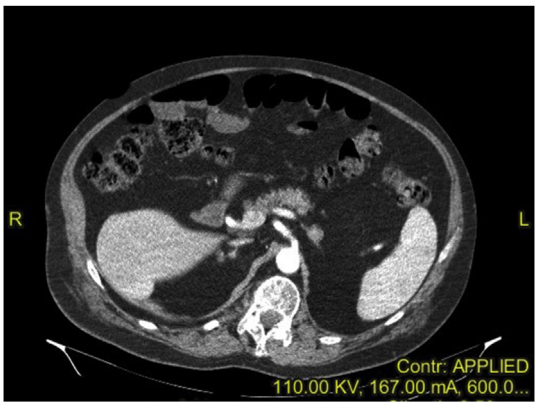

Fig. 2. Left adrenal on CT-scan.

Treatment options were considered including medical management with lifelong steroid replacement therapy.

Patient's perioperative risk was increased by coronary artery disease (myocardial infarction 20 years prior) with preserved systolic function and second-degree diastolic dysfunction, residual dysphonia (present at discharge from gamma-knife surgery), morbid obesity (body mass index: 32.8 $kg\,m^{-2}$) and obstructive sleep apnea syndrome (OSAS) as diagnosed with sleep monitoring.

After a multidisciplinary team (MDT) work-up involving a endocrinologist, a urologist and a radiologist, we came to the decision to treat the patient with bilateral laparoscopic adrenalectomy. A formal consent was obtained for surgical management.

Anesthesia was induced with propofol 1.5 mg/kg and fentanyl 3 mcg/kg; lidocaine 2%, and tracheal intubation was facilitated with cisatracurium 2 mg/kg, after which sevoflurane and remifentanil were used for maintenance. The highest blood pressure recorded during induction was 132/79 mmHg.

Surgical technique. The patient was placed in a left lateral decubitus position. After the first 12mm trocar placement for the camera, a 12 mmHg pneumoperitoneum was established. Standard adrenalectomy was perfomed.

The patient was then placed into a right lateral decubitus position for right adrenalectomy.

Both adrenal glands were removed with an endocath bag through the umbilical port, 5cm extended for specimens retrieval.

After resection of the glands, 100 mg of hydrocortisone were administered in bolus, and a norepinephrine infusion was used to maintain mean arterial blood pressure $\geq$60 mmHg; maximum infusion rate was 0.07 μg/kg/min (ideal body weight), and the patient was weaned from the infusion by the time surgery ended.

The patient was transferred to the surgical intensive care unit for monitoring during the day of surgery, where he stayed for 16 hours, with an unremarkable course except for slight elevation of bilirubin and liver enzymes, which resolved spontaneously the next day. He was transferred to the surgical ward on the morning of postoperative day 1. During post-operative time, no complications occurred. Drainages were removed on day 2. The patient was discharged home on postoperative day 4 after starting steroid replacement. Histopathologic analysis confirmed macronodular adrenal hyperplasia.

After a 6 months follow-up the patient had a weight drop of 5 kg. Glucose control, blood pressure and patient's quality of life were significantly improved. The patient is currently under follow-up visits and is on cortone-acetate 37,5 mg OD.

2. Discussion

The first laparoscopic adrenalectomy was described by Gagner et al., in 1992 [2]. Since then, the laparoscopic approach has become more and more widespread and today it is considered the gold standard for adrenalectomy. Open adrenalectomy would require a large incision to gain access to a relatively small gland, while laparoscopy shows many advantages, such as a shorter hospital stay, reduced morbidity, decreased analgesic requirement, smaller wound, reduced intraoperative blood loss, and a rapid recovery of patient's strength.

The laparoscopic approach for adrenalectomy has been widely explored and is now considered safe and effective even in case of large masses [3–5]. While there is not common agreement in literature about the definition of “large” adrenal masses, ranging from $\geq$3.5 to $\geq$8 cm among different authors, a contraindication remains absolute: the involvement of surrounding tissue or adrenal and caval vein by a malignant adrenal lesion.

In our case CT-scan showed small adrenal masses, 3 and 4 cm respectively, thus suggesting the laparoscopic approach to be feasible and safe.

Bilateral adrenal lesions requiring surgical management are not common. Cushing's disease is ACTH-dependent and is accompanied by bilateral diffuse adreno-cortical hyperplasia, which is usually treated by trans-sphenoid surgery or bilateral total adrenalectomy [6]. In these cases, laparoscopic bilateral adrenalectomy has been described and can offer definitive treatment with low morbidity and less complications than open surgery [7,8].

The first series of patients treated with bilateral synchronous laparoscopic adrenalectomy was carried out by Hsu and Gill in 2002, showing that the procedure results in minimal postoperative morbidity [9].

Clow et al. reported the outcome of laparoscopic synchronous bilateral adrenalectomy in 68 patients diagnosed with ACTH-dependent Cushing's syndrome, most of whom were resistant to previous pituitary surgery [10]. Similarly, Takata et al. reported the results of laparoscopic simultaneous bilateral adrenalectomy with Cushing's disease and pheochromocytoma, and concluded that the procedure was safe and effective [11].

Our case was particularly interesting, due to his wide preoperative comorbidity. He suffered from other endocrine pathologies, like toxic multinodular goitre; he was previously treated for neurofibromatosis type 2 and was cardiopathic with severe concentric hypertrophy and diastolic dysfunction. Last but not least, he was morbidly obese (a condition making the laparoscopic approach more challenging) and affected by OSAS (a well known risk factor complicating anesthesiological management).

Despite the technical challenges due to morbid obesity, a laparoscopic approach was granted, though, as this has been associated with reduced length of stay and blood loss [12] - the latter factor being extremely important in this case, where even a minor DVT episode might have led to a catastrophic occlusion of the caval filter, and in whom immediate restoration of systemic anticoagulation was indicated. Reduced surgical wound size, and postoperative pain, may also translate in lower opioid consumption, which should in turn reduce the short-

term risk for respiratory and gastrointestinal complications. Although these outcome parameters have not specifically been addressed in adrenalectomy trials, experience from live kidney donor surgery shows significantly lower postoperative pain and improved respiratory function [13,14]. A minimally invasive approach is, thus, very likely to improve the overall outcome of bilateral adrenalectomy, also in terms of a reduction in respiratory complications—which is particularly important in patients with OSAS, who have roughly double the risk of postoperative complications [15]. Additionally, early mobilization of a patient at high risk for recurrent DVTs was of paramount importance, and such goal is more likely to occur with less invasive surgery.

4. Conclusion

Although medical management of Cushing's syndrome in this patient might have been feasible, life-long therapy starting at the age of 51 was deemed impractical in terms of risk of side effects and difficulties in maintaining adequate hormonal balance. Once the decision was made, together with our patient, to proceed with surgery, we opted for a bilateral laparoscopic approach as part of a balanced enhanced recovery program which resulted in uneventful discharge after 4 days. We postulate that the ability to mitigate our patient's comorbidities and perioperative risk factors with a bilateral laparoscopic resection was as important as the more surgery-specific advantages (such as lower blood loss).

Ethical approval

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Author contribution

Every Author contributed equally.

Conflicts of interest

No conflict of interest.

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Not applicable.

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Guarantor

Mr. Francesco Ziglioli.

Patient consent

Written informed consent was obtained from the patient for the publication of this case report and accompanying images.

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