

Bilateral Laparoscopic Adrenalectomy in a Pregnant Woman with Cushing's Syndrome

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INTRODUCTION

Cushing's syndrome (CS) occurs rarely during pregnancy.⁽¹⁾ Surgical treatment is the principal therapy for CS in pregnancy, with medical treatments constituting the second choice. The most common cause of CS in pregnancy is adrenal gland adenoma that may be treated by unilateral adrenalectomy during gestation.^(2,3)

Herein we present a 31 years old pregnant woman with CS who underwent bilateral laparoscopic adrenalectomy in her 18th week of pregnancy. Our Medline search revealed that this is the first "bilateral" laparoscopic adrenalectomy during pregnancy which has been reported.

CASE REPORT

A 31-year old primigravid woman was referred to our clinic at 4 months gestation following the diagnosis of CS from about three months ago. She has been treated medically but her blood pressure was not well controlled. The patient also had a history of cardiac ablation because of paroxysmal supraventricular tachycardia about 4 months ago when she was unaware of her pregnancy.

Her laboratory data showed urine free cortisol level of 730 $\mu\text{g}/24\text{hr}$ (normal range, 10-100) and her late-night salivary cortisol level was 60 nmol/L (normal range, 6.2-19.4). Ultrasonography showed increased size of adrenals with an 18 weeks normal fetus (**Figure 1**). Abdominal magnetic resonance imaging (MRI) showed adrenal hyperplasia (**Figure 2**) but brain MRI was normal. We decided to perform bilateral laparoscopic adrenalectomy. Laparoscopy was done under general anesthesia with anterior transperitoneal approach using three working trocars. After extraction of left adrenal gland (**Figure 3**) the position was changed to right lateral decubitus and right adrenal gland was similarly excised successfully (**Figure 4**). The time of surgery was about 4.5 hours. Post-operative view of the abdomen is shown in **Figure 5**. The operation was uneventful and the patient and fetus were well during post-operative period. The patient was discharged after 4 days with oral medications.

After surgery baseline serum cortisol level decreased from 60 to 2.8 nmol/L, and 24-hour urine cortisol decreased from 730 to 29 μg . After 3 months the patient underwent cesarean section because of fetoplacental abnormality. A preterm infant with intrauterine growth retardation (IUGR) without any malformation was born. The patient was discharged without any complication but her infant was remained under observation in neonatal intensive care unit (NICU) for

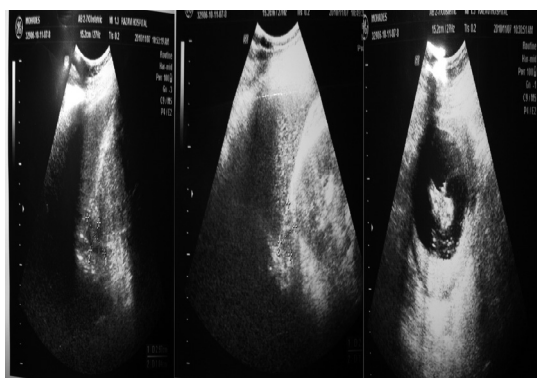


Figure 1. Ultrasonography of the patient.

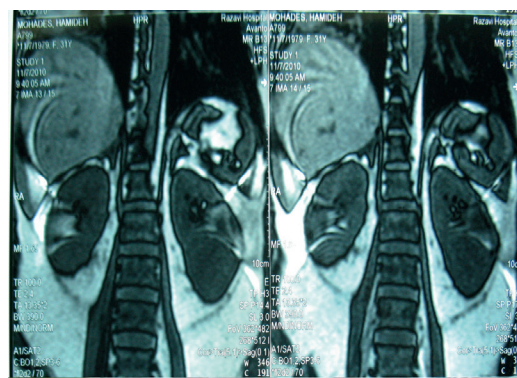


Figure 2. Magnetic resonance imaging of the patient.

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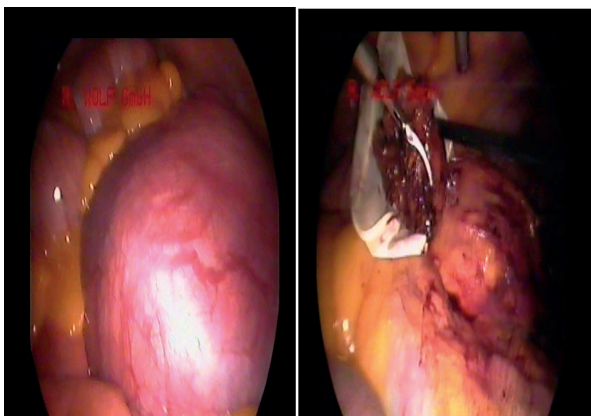


Figure 3. Laparoscopic views. Left: The pregnant uterus. Right: The adrenal during excision.

40 days. The baby had good condition after discharge and remained normal during his two years follow up.

DISCUSSION

Pregnancy is rare in women with CS, with fewer than 150 cases reported in the literature. It is because that the hyperandrogenism and hypercortisolism status during pregnancy suppress pituitary secretion of gonadotropins.^(4,5) However, because CS results in increased fetal and maternal complications, its early diagnosis and treatment are critical.^(1,2) The etiology of CS in pregnant women is different from that in of non-pregnant women. Adrenal adenomas cause approximately 40-50% and 17-29% of CS in pregnant and non-pregnant women, respectively.^(4,6,7) In contrast, Cushing disease is less common in pregnancy; with rates of 63-72% in the general population compared with 33% in pregnant women.^(4,6) The etiology of CS in our case was bilateral adrenal hyperplasia. There are many overlapping features between normal pregnancy and CS, so the clinical diagnosis of CS in pregnancy may be difficult and unfortunately is often not detected until 12-26 weeks of gestation.^(4,6,7) The biochemical diagnosis of CS in pregnancy is difficult because of the normal hypercortisolism during pregnancy.⁽⁶⁾ Adrenal

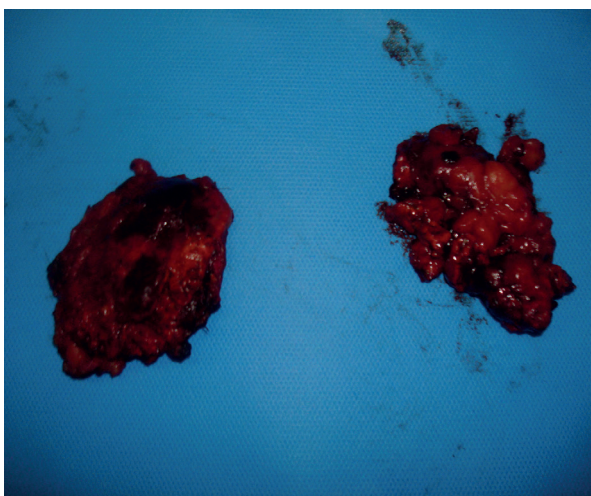


Figure 4. Adrenal specimens after excision.

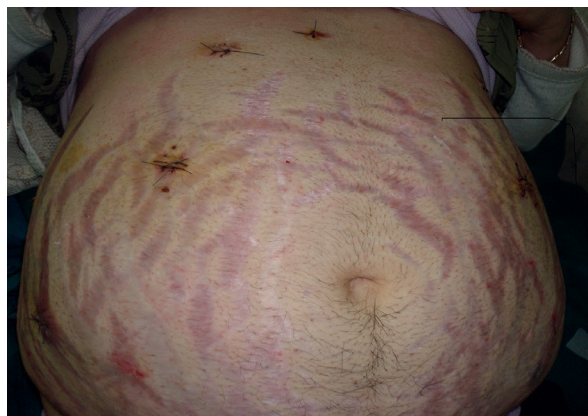


Figure 5. Abdomen at the end of surgery.

ultrasonography or MRI can be safely performed during pregnancy for detection of adrenal tumor. MRI can also be useful in locating pituitary tumors.⁽³⁾

When CS is diagnosed during pregnancy, therapeutic options depend on the underlying etiology, including, surgical treatment, conservative management, medical treatment, and delay of surgery until after delivery.⁽⁵⁾ When contemplating surgical treatment for a pregnant patient with suspected adrenocortical adenoma, the surgical approach and the optimal time for surgery need to be determined. Surgery is the treatment of choice for CS in pregnancy, except perhaps late in the third trimester, with medical treatment being a second choice. There is no rationale for supportive treatment alone.⁽⁴⁾ The commonest cause of CS in pregnancy, adrenal gland adenoma, may be treated by unilateral adrenalectomy during gestation. Open or laparoscopic methods have been performed in practice. The end of the first trimester and the first half of the second trimester are considered the best time for surgery. In the third trimester, conservative treatment and early delivery are preferred. However Aishima and colleagues and Sammoura and colleagues successfully treated their patients with CS at this gestational stage by retroperitoneal laparoscopic adrenalectomy.^(5,8) We found 24 cases of adrenalectomy in literature due to CS caused by adrenocortical adenoma in pregnancy, but we didn't find a case of bilateral laparoscopic adrenalectomy in a pregnant woman. In our case, laparoscopy was done by transperitoneal approach. Although the patient was obese with a history of severe hypertension and cardiac ablation, she didn't have any complication perioperatively.

CONCLUSION

We demonstrated that bilateral laparoscopic adrenalectomy is possible during pregnancy and may be considered safe and minimally invasive in selected patients.

CONFLICT OF INTEREST

None declared.

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