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Case report
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A RARE CASE OF ADRENAL CAVERNOUS HEMANGIOMA

REDAK SLUČAJ NADBUBREŽNOG KAVERNOZNOG HEMANGIOMA

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Summary
Introduction. Adrenal cavernous hemangiomas are extremely rare non-functioning benign tumors. The majority of adrenal cavernous hemangiomas are diagnosed incidentally after surgery by histopathological examination. **Case Report.** We report a clinical case of a 57-year-old woman with adrenal cavernous hemangioma. On admission, the patient's adrenal-related hormones were in the reference range, so adrenal endocrine dysfunction was ruled out. The computed tomography scan revealed a well-circumscribed, round, heterogeneous right adrenal mass (32.3 x 55.4 mm). Iodinated contrast enhanced abdominal computed tomography showed a slight inhomogeneity. In this case, preoperative radiologic findings and absence of signs of local invasion indicated laparoscopic adrenalectomy. The patient underwent right transperitoneal adrenalectomy. Microscopic evaluation showed a sinusoidal dilatation and fibrotic septa, so postoperative diagnosis of adrenal cavernous hemangioma was made. **Conclusion.** In summary, we reported a case of an incidentally discovered non-functioning adrenal cavernous hemangioma treated by laparoscopic surgery. The diagnosis of adrenal cavernous hemangioma may be challenging, and it is commonly made after surgery, since it is frequently confirmed by histopathological examination.
Key words: Hemangioma, Cavernous; Adrenal Gland Neoplasms; Laparoscopy; Diagnosis; Incidental Findings

Sažetak
Uvod. Kavernozni hemangiomi nadbubrega su izuzetno retki nefunkcionalni benigni tumori. Većina kavernoznih hemangioma nadbubrega dijagnostikovana je nakon operacije histopatološkim pregledom. **Prikaz slučaja.** Prikazujemo klinički slučaj kavernoznog hemangioma nadbubrežne žlezde kod 57-godišnje žene. Pri prijemu, hormoni nadbubrežne žlezde kod pacijentkinje bili su u referentnom opsegu, tako da smo isključili bilo kakvu adrenalnu endokrinu disfunkciju. Skeniranjem kompjuterizovanom tomografijom videla se jasno ograničena, okrugla, heterogena masa u desnoj adrenalnoj žlezdi (32,3 x 55,4 mm). Kompjuterizovana tomografija abdomena sa jodnim kontrastom pokazala je neznatnu nehomogenost. U ovom slučaju preoperativni radiološki nalazi i nedostatak znakova lokalne invazije, naveli su nas da izvedemo laparoskopsku adrenalektomiju kako bismo uspostavili konačnu dijagnozu. Pacijentkinja je podvrgnuta desnoj adrenalektomiji transperitonealnog lumbalnog pristupa. Mikroskopska evaluacija pokazala je sinusoidnu dilataciju i fibrozne lezije sa postoperativnom dijagnozom kavernoznog hemangioma nadbubrežne žlezde. **Zaključak.** Ukratko, prikazali smo slučaj, slučajno otkriven nefunkcionalni adrenalni hemangiom tretiran laparoskopskim pristupom. Dijagnoza kavernoznog hemangioma najčešća je nakon operacije, a najčešće se potvrđuje histopatološkim pregledom.
Ključne reči: kavernozni hemangiom; neoplazme nadbubrežne žlezde; laparoskopija; dijagnoza; slučajni nalaz

Introduction

Adrenal masses are discovered with increasing frequency due to widespread use of radiological imaging techniques. It is estimated that adrenal masses are incidental findings in 1% to 5% of all abdominal computed tomography (CT) scans [1]. Adrenal cavernous hemangiomas are extremely rare non-functioning benign tumors [2]. The majority of adrenal cavernous hemangiomas are diagnosed after surgery by histopathological examination. Approximately 60 surgical cases have been reported in the literature so far [2].

The current report presents a case of an adrenal cavernous hemangioma which was confirmed histologically after laparoscopic surgery.

Case Report

We report a clinical case of adrenal cavernous hemangioma in a 57-year-old woman. An incidental right adrenal mass was discovered on ultrasonography when she visited a nephrologist due to a right flank discomfort. Her past medical history was positive for essential hypertension in the past year which was treated by 20 mg lisinopril + 12.5 mg hydrochlorothiazide twice daily. She also had type 2 diabetes mellitus in the past two years, and it was treated by glibenclamide 5 mg once daily in the morning. Physical examination did not reveal any abnormalities. The patient's blood pressure was 140/90 mmHg, with a pulse rate of 78 beats/min. On admis-

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Abbreviations
CT – computed tomography
MRI – Magnetic Resonance Imagining
H&E – Hematoxylin and Eosin

sion, laboratory findings **(Table 1)** and adrenal-related hormones and other hormones **(Table 2)** were in the reference range, so any adrenal endocrine dysfunction was ruled out. The abdominal CT scan revealed a well circumscribed, heterogeneous right adrenal mass (32.3 x 55.4 mm) **(Figure 1)**. Iodinated contrast enhanced abdominal CT showed a slight inhomogeneity. In this patient, abdominal magnetic resonance imaging (MRI) was not performed. The preoperative radiologic findings and the absence of signs of local invasion indicated laparoscopic adrenalectomy in order to establish the final diagnosis. The perioperative blood glucose level was 5.01 mmol/l (reference range: 3. 5 – 6.1 mmol/l) well controlled with glibenclamide. The antihypertensive therapy was continued on the day of surgery. The patient underwent a right transperitoneal adrenalectomy. Intraoperatively, an encapsulated adrenal mass without local invasion was found. The total operating time was 160 min, without any intraoperative or postoperative complications. The patient was discharged on postoperative day 6. Microscopic evaluation showed sinusoidal dilatation and fibrotic septa and a postoperative diagnosis of adrenal cavernous hemangioma was made **(Figure 2)**. Positive cluster of differentiation 31 and cluster of differentiation 34 immunostaining demonstrated a tumor of endothelial nature that supported the diagnosis [3].

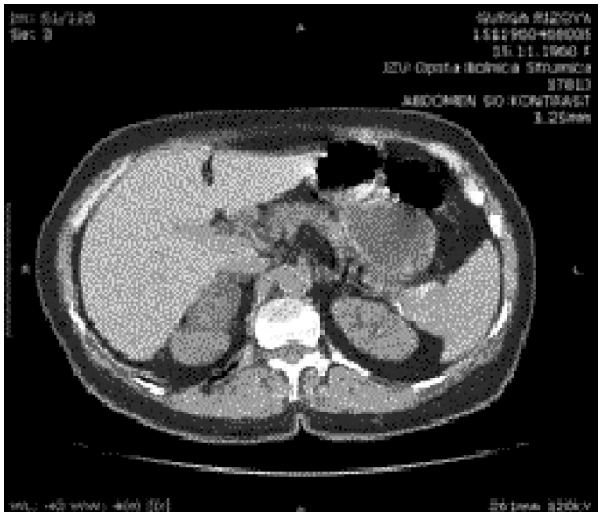

Figure 1. Abdominal CT scan revealed heterogenous right adrenal mass with regular margins
Slika 1. *Kompjuterizovana tomografija abdomena otkrila je heterogenu desnu nadbubrežni masu sa regularnim marginama*

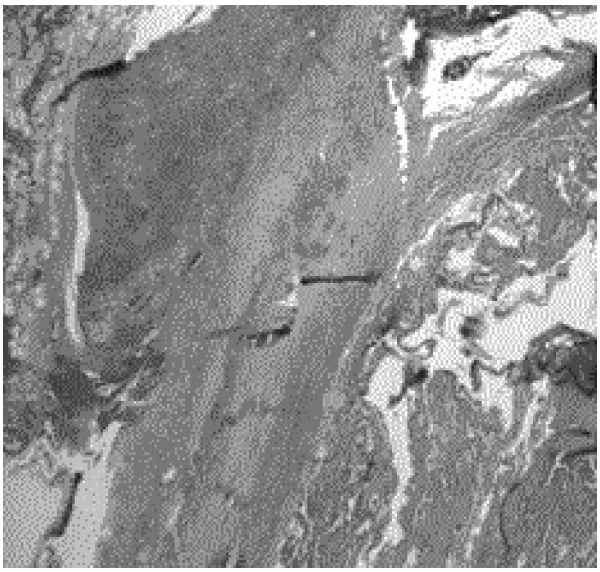

Figure 2. Histopatology. Thick walled vascular cavities are present adjancent to the adrenal cortex. (H&E stain original magnifaction 13x)
Slika 2. *Histopatologija. Vaskularne šupljine sa debelim udovima su prisutne u korteksu nadbubrežne žlezde (H&E obojeno originalno uvećanje 13x)*

Discussion

Cavernous hemangiomas of the adrenal gland are rare, benign and endocrinologically inactive tumors. The pathogenesis of these tumors is unclear; they are probably congenital, with involvement of hereditary factors and ectasia of blood vessels [3]. They are usually unilateral and appear in the sixth or seventh decade of life. The majority of tumors are asymptomatic lesions, but if they are large, the symptoms include flank pain and a palpable mass, or they may present with hypovolemic

Table 1. Laboratory test results
Tabela 1. *Laboratorijski nalazi*

Test/*Test*	Result/*Rezultat*	Reference range/*Referentne vrednosti*
WBC (white blood cells) x10^9/L/*Leukociti*	7.4	4.00 - 9.00
Hematocrit/*Hematokrit (rv)*	0.440	0.37 - 0.54
PLT (platelet count) x10^9/L/*Trombociti*	271	150 - 450
Glucose (mmol/l)/*Glukoza*	5.01	3.5 - 6.5
Urea (mmol/l)/*Urea*	4.5	2.7 - 7.8
Creatinine (umol/l)/*Kreatinin*	83	45 - 109
C-reactive protein mg/L/*C-reaktivni protein*	4	< 6

Table 2. Levels of the patient's adrenal hormones, other hormones, and electrolytes during the preoperative period
Tabela 2. *Vrednosti adrenalnih hormona, drugih hormona i elektrolita kod pacijentkinje u preoperativnom periodu*

Test *Test*	Result *Rezultat*	Reference range *Referente vrednosti*
Morning cortisol levels/*Jutarnji kortizol u serumu (nmol/l)*	120.96	55 - 690
Cortisol levels at night/*Večernji kortizol u serumu (nmol/l)*	20 h 145.5	55 - 690
Midnight serum cortisol/*Ponoćni kortizol u serumu (nmol/l)*	24 h 87.8	55 - 690
Adrenocorticotropic hormone/*Adrenokortikotropni hormon (ρg/ml)*	21	< 46
Aldosterone/*Aldosteron (ng/ml)*	11.2	5 - 14.5
Calcitonin/*Kalcitonin (ρg/ml)*	< 3	< 5.8
Sodium/*Natrijum (mmol/l)*	140	137 - 145
Potassium/*Kalijum (mmol/l)*	4	3.8 - 5.5
Ionized calcium test/Jonizovani *kalcijum (mmol/l)*	1.26	1 - 1.30
Dexamethasone suppression test *Test prekonoćne supresije deksametazonom(cortisol/kortizol nmol/l)*	42	55 - 690
Urinary metanephrines/*Metanefrini u urinu* Metanephrines/*Metanefrini (μg/24 h)*	68	< 90
Urinary normetanephrines/*Normetanefrini u urinu* Normetanephrines/*Normetanefrini (μg/24 h)*	134	< 180

shock caused by a spontaneous rupture. Adrenal hemangiomas are most commonly non-functioning tumors, and only three cases of functioning adrenal hemangiomas have been reported to date [4].

Adrenal masses are usually identified by imaging techniques performed for other reasons. The differential diagnosis of incidental adrenal masses includes: adrenal adenoma, adrenal cortical carcinoma, metastatic cancer, pheochromocytoma, adrenal cyst, myelolipoma, hematoma, ganglioneuroma and cavernous hemangioma. A CT scan finding of adrenal cavernous hemangioma includes a hypodense, heterogeneous lesion with calcifications. Spotty calcifications throughout the tumor are probably due to phleboliths in dilated vascular spaces [5]. Although nonspecific, abdominal MRI may show hyperintensity on T2-weighted images and a focal hyperintensity on T1-weighted images [6].

On histopathological examination, adrenal cavernous hemangioma is located in the adrenal cortex and consists of multiple dilated vascular cavities lined by a single layer of vascular endothelium surrounded by a collagenous wall [7]. The main indications for surgery of these adrenal masses are to relieve mass-effect-type symptoms, to exclude malignancy, and to treat complications such as spontaneous bleeding [8].

Conclusion

In summary, we reported a case of an incidentally discovered nonfunctionine adrenal cavernous hemangioma treated by laparoscopic surgery. The diagnosis of adrenal cavernous hemangioma may be challenging and it most often occurs after surgery, since it is frequently confirmed by histopathological examination. Surgical excision may be achieved by laparoscopic adrenalectomy, as a standard care for small to medium sized benign adrenal tumors (up to 6 – 7 cm in diameter).

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