



Case series

Acute adrenal insufficiency due to adrenal hemorrhage complicating colorectal surgery: Report of two cases and correlation with the antiphospholipid antibody syndrome

Angeliki Kolinioti^{a,*}, Michalis Tsimaras^b, Georgios Stravodimos^b, Vasileios Kompodoros^b

^a 3d Surgical Department, Evaggelismos General Hospital, Athens, Greece

^b 4th Surgical Department, Evaggelismos General Hospital, Athens, Greece

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ABSTRACT

INTRODUCTION: Spontaneous bilateral adrenal hemorrhage or hemorrhagic necrosis due to adrenal vein thrombosis is an uncommon condition that may lead to acute adrenal insufficiency and death. The objective of this report is to enhance recognition of this potentially fatal disorder in surgical patients.

PRESENTATION OF CASES: We present two cases of acute adrenal insufficiency due to bilateral adrenal hemorrhage associated with primary antiphospholipid antibody syndrome (APS). Both cases occurred in the early postoperative period after major colorectal surgery. Major vein thrombosis, abdominal pain, anorexia, asthenia, lethargy and an unexplained drop in patient's hemoglobin without evidence of sepsis were the principal symptoms and signs that, with a high index of suspicion, led to the correct diagnosis. **DISCUSSION:** Antiphospholipid syndrome is an acquired thrombophilia caused by circulating antibodies against a heterologous group of phospholipids. Recent literature has identified a causative relation between APS and primary adrenal insufficiency (AI), identifying it as its most common endocrine manifestation. Surgeries along with inflammation or hormones have been identified as precipitating factors. Spontaneous haemorrhagic infarction of the adrenal glands has been observed in patients with APS in the postoperative period during anticoagulant treatment. Signs and symptoms are non-specific and are easily confused with those of the underlying condition.

CONCLUSIONS: Early recognition and prompt treatment of adrenal insufficiency due to APS in surgical patient is of vital importance. Patients correctly diagnosed and treated that survive the critical phase have a better prognosis regarding restoration of adrenal function.

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1. Introduction

We present two surgical cases complicated with acute adrenal insufficiency, due to bilateral adrenal hemorrhage associated with primary antiphospholipid antibody syndrome (APS). Cases are reported in line with the SCARE criteria [1]. The objective of our report is to enhance prompt diagnosis of this potentially fatal dis-

order by analyzing its clinical course, diagnostic algorithm and causative factors.

2. Case report

2.1. Case 1

A 65 year old female with unremarkable past medical history was admitted to our clinic as an emergency with large bowel obstruction. CT scan of the abdomen showed distended loops of small and large bowel with a transition point at the level of the sigmoid colon. After an enema administration per rectum to clean the distal bowel, the patient underwent a flexible sigmoidoscopy which confirmed the presence of an obstructing carcinoma 25 cm above the dentate line. Blood results revealed anemia (Ht = 29%) and an elevated white cell count (12500/mm³). Electrolytes were nor-

Abbreviations: APS, antiphospholipid antibody syndrome; AI, adrenal insufficiency; ACTH, adrenocorticotropic hormone; DVT, deep vein thrombosis; CT, computerized tomography; LA, lupus anticoagulant; aCL, anticardiolipin antibody; anti-(2)GPI, anti-β(2)-glycoprotein I antibodies.

* Corresponding author at: 3d Surgical Department, Evaggelismos Hospital of Athens, 45 - 47 Ipsilantou Street, 106 76, Athens, Greece.

E-mail address: akolinioti@gmail.com (A. Kolinioti).

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Fig. 1. Abdominal CT scan of case1 demonstrating a mixed attenuation mass in both adrenal glands. There is some preservation of normal adrenal enhancement in the periphery.

mal and no clotting abnormalities were identified preoperatively (Fig. 1).

Colonic stenting is not commonly performed in our Institution for resectable colonic neoplasms. Therefore, the patient proceeded to an exploratory laparotomy. Findings were of a sigmoid tumor with no evidence of metastatic disease. In the view of an obstructed bowel, an abdominal colectomy with ileorectal anastomosis was performed. Postoperative course was uneventful until the fourth postoperative day when the patient developed a fever (37.8 C) with fatigue, lethargy and hypotension combined with hyponatraemia (120mEq/l) and relative hyperkalemia (4.7mEq/l). There was clinical evidence of an acute deep venous thrombosis of the left subclavian, left jugularis internal and external, confirmed on ultrasound, even though the patient received DVT prophylaxis with low molecular weight heparin from postoperative day one. A CT scan of the abdomen was performed at that point to exclude abdominal pathology. It showed bilateral enlargement of the adrenal glands (6 cm on the right and 4 cm on the left) with evidence of recent hemorrhage. Plasma cortisol was calculated and found to be very low (5 µg/dl) with an ACTH of 290 pg/ml. Further laboratory investigation with ELISA showed elevated levels of anticardiolipin antibodies – IgG 22.6 GPL (ref. 0–20 GPL) and IgM 35.5 MPL (ref 0–20 MPL).

The patient rapidly improved with the administration of a pulse dose of steroids. She also received anticoagulation treatment, initially with heparin and then with coumarin derivatives. Six months later, a follow-up CT scan showed normal adrenal glands. Steroids were successfully tapered, but anticoagulation treatment was continued for life (Fig. 2).

2.2. Case 2

A male 72 year old patient was referred to our Colorectal Clinic with a diagnosis of intractable ulcerative colitis without extra-colonic manifestations. The patient received 40 mg prednisolone per day at home and continued to be severely symptomatic. He proceeded to have a total colectomy with ileoanal anastomosis, J pouch and prophylactic loop ileostomy. The operation was uneventful and the patient received appropriate DVT prophylaxis with low molecular weight heparin. Stress dose steroids were administered on induction of anaesthesia and were continued accordingly, as

the patient was previously on long term steroid treatment for his ulcerative colitis.

He had a good recovery and resumed oral intake on the fourth postoperative day. On the fifth postoperative day, he developed acute deep venous thrombosis of the left lower limb, confirmed on Doppler ultrasound, and pulmonary embolism on CT Pulmonary Angiography. He was initially treated with heparin (30,000 IU i.v.) and continued with coumarin derivatives (Acenocumarol 2 mg/ day p.o.).

When steroid tapering was attempted postoperatively, the patient presented with clinical manifestations of adrenal insufficiency. He developed anorexia, lethargy, hypotension, hyponatraemia; fever (T 39 C). A drop in the patient's haemoglobin was noted (from 12 g/dL to 9.7 g/dL) without evidence of blood loss, haemolysis or sepsis. A CT scan of the abdomen showed enlargement of both adrenal glands with evidence of recent haemorrhage. Further laboratory investigations identified positive anticardiolipin antibodies (IgG 23.6 GPL, IgM 67.2 MPL).

His condition improved with hydrocortisone treatment (40 mg/day p.o.). Two months postoperatively, another attempt of hydrocortisone tapering led to a relapse of adrenal insufficiency. The patient resumed his previous hydrocortisone regime, until six months later when, after a successful tapering, hydrocortisone was stopped. The adrenal glands appeared normal on follow up imaging. Anticoagulation treatment with coumarin anticoagulants was continued for life (Fig. 3).

3. Discussion

Bilateral adrenal haemorrhage is a rare but potentially fatal condition, leading to the development of acute adrenal insufficiency, when at least 90% of the glands are injured. The adrenal gland has a distinct vascular anatomy; a high flow arterial network with an abrupt transition to a single vein which renders the gland vulnerable to haemorrhagic events.

The majority of post mortem studies indicate that the sequence of events is a thrombosis-mediated adrenal venous occlusion causing edema of the gland and secondary obstruction of the arterial supply resulting in a hemorrhagic infarction [2]. Espinosa et al retrospectively reviewed 86 cases of APS related adrenal haemorrhage. In 6 out of the 22 cases (27%) where histopathology report

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