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Posterior reversible encephalopathy syndrome (PRES) in mesenteric leiomyosarcoma: A case report

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ABSTRACT

BACKGROUND: Posterior reversible encephalopathy syndrome (PRES) is a syndrome characterized by headache, confusion, visual loss and seizures. Many factors influence the appearance of this syndrome, predominantly eclampsia, certain medical treatments and malignant hypertension. Diagnosed by typical transient lesions on magnetic resonance imaging.

CASE REPORT: We present a case of mesenteric leiomyosarcoma in a 52 year old woman, who had severe headache, abdominal heaviness, and hypertension. Investigations revealed a mesenteric mass and a **P**osterior **R**eversible **E**ncephalopathy **S**yndrome features on brain MRI, suggesting renin secretion by the tumor, causing the patient's symptoms.

CONCLUSION: Patient's symptoms disappeared after resection of the tumor, suggesting a renin production cessation.

1. Introduction

Posterior reversible encephalopathy syndrome (PRES), also known as reversible posterior leukoencephalopathy syndrome (RPLS), is a syndrome characterized by headache, confusion, visual loss and seizures. Many factors influence the appearance of this syndrome, predominantly eclampsia, certain medical treatments and malignant hypertension. The symptoms can resolve spontaneously after a period of time, although visual sequelae have been reported [2,3].

The present case details a patient with mesenteric leiomyosarcoma and liver metastases who presented with headache, hypertension and PRES. Resection of her primary and metastatic disease led to resolution of PRES and hypertension.

Our case has been reported in line with the SCARE criteria [1].

2. Case presentation

A 52 year old female presented abdominal pain and severe headaches with visual disturbances. Headaches were refractory to pain medication. Initial imaging revealed an abdominal mass and liver lesion, and surgery was recommended. She was referred to our clinic where we noted severe hypertension reaching peaks of 190/110 mmHg associated with unremitting headache. The headache was described as tension-like, starting bilaterally in the occipital region and radiating to all other regions while progressive in severity over the course of two weeks prior to presentation at our clinic. These episodes were accompanied by nausea and vomiting.

Neurological examination was non-focal. Physical examination revealed a palpable left-central abdominal mass, freely mobile in the direction perpendicular to the mesentery. The patient reported that she initially noted the mass three years prior. She denied gastrointestinal symptoms. Laboratory workup was unremarkable with normal electrolytes, coagulation, liver and renal function. The patient's history was notable for resection of a uterine leiomyoma 14 years prior; she had four pregnancies all resulting in healthy births (children aged 16–22 years).

She was immediately treated with 5 mg bisoprolol and 300 mg/10 mg irbesartan/amlodipine and underwent brain magnetic resonance imaging and multiphasic computed tomography (CT) of the abdomen and pelvis.

MRI revealed bilateral symmetrical occipital cortical-subcortical high T2/FLAIR signal intensity highly suggestive of PRES (Fig. 1). Abdominal computed tomography revealed a 7.3 × 5.8 cm well-circumscribed mesenteric mass located on the left believed to be related to the mesentery showing heterogeneous significant enhancement with hypodense areas. No invasion or mass effect for the adjacent structuresand a 4 cm solitary lesion of the left liver consistent with metastasis from the mesenteric lesion (Fig. 2).

Medical therapy controlling hypertension led to complete resolution of headaches and repeat MRI was normal. Thus we proceeded to surgery for the mesenteric and liver lesions. Operative findings included a lesion situated within the sigmoid mesocolon

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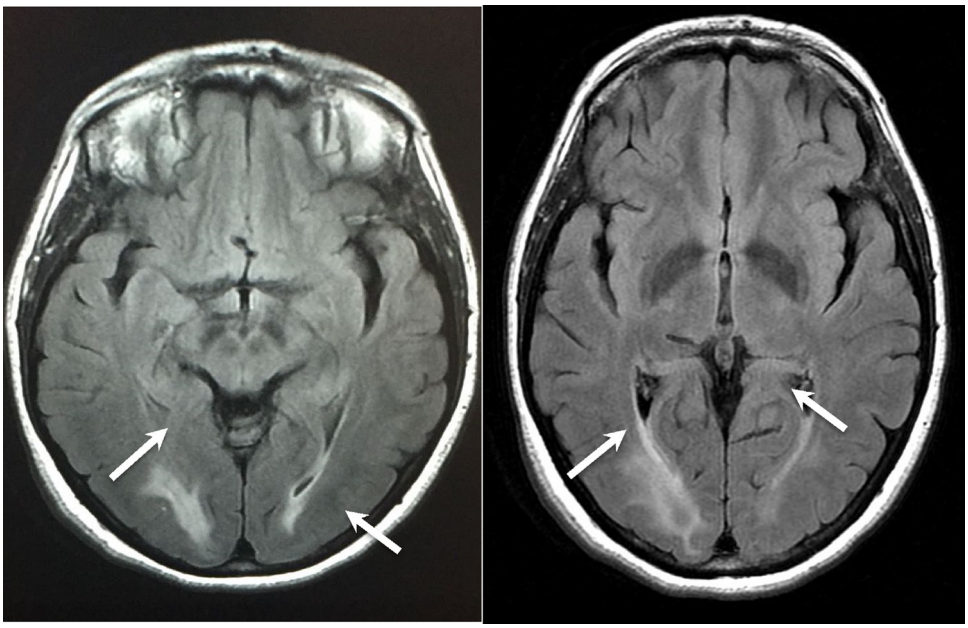

Fig. 1. Brain MRI. Arrows indicate Bilateral symmetrical occipital cortico-subcortical high T2/FLAIR signal intensity suggesting a vasogenic oedema within the occipital and parietal regions most likely relating to the posterior cerebral artery supply This confirm Posterior Reversible Encephalopathy Syndrome (PRES).

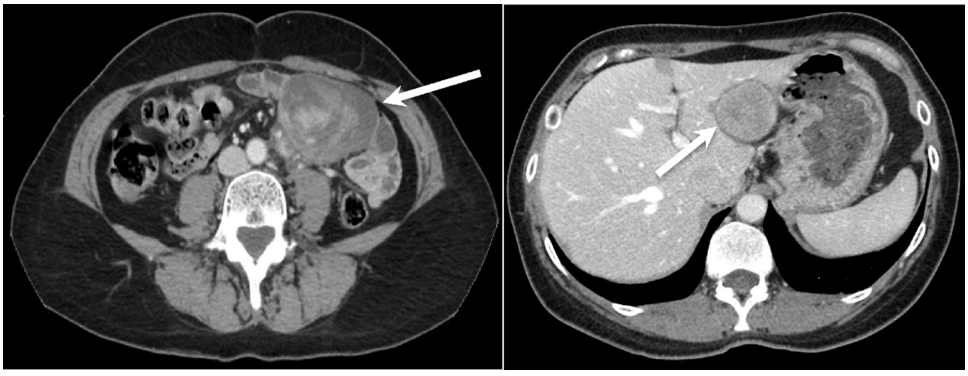

Fig. 2. Computed tomography: Left panel, mesenteric mass with mixed hyper- and hypo- attenuating areas on arterial phase imaging; Right panel: similar appearing lesion in segment II liver.

approximately at the level of the iliac bifurcation; all vessels to the lesion emerged from the mesocolon, none from the retroperitoneum proving the mesenteric origin. It was not associated with the colon per se. The lesion and mesentery lay laterally displacing retroperitoneal structures including the left ureter laterally. Complete resection was possible without sigmoid resection.

At the same laparotomy, intraoperative ultrasound revealed 2 left lateral liver lesions, the known larger lesion in segment III and an adjacent smaller lesion in segment II. Lateral bisegmentectomy was completed with widely negative margins. Intraoperative ultrasound revealed no other liver lesions. The character of the liver was normal. There was no evidence of ascites, adenopathy or peritoneal disease. The operation was completed in 2 h, with less than 20 cc of blood loss (Fig. 3).

The postoperative course was remarkable for resolution of hypertension. In fact, antihypertensive medications were slowly discontinued. Headache did not recur in the immediate perioperative period. The patient was discharged from the hospital the fifth postoperative day. Multidisciplinary consensus was that absent any residual disease, adjuvant therapy was not appropriate.

Pathology revealed a left mesenteric mass: 10 × 7 × 6,5 cm mass, weighing 217 g, well circumscribed with smooth outer aspect, serially cut, presence of some whitish fibrillary areas with predominant shiny grayish myxoid pattern with multifocally dilated congestive blood vessels with hemmorhagic spaces containing blood clots. and liver 12 × 11 cm in major dimensions and 2,5 cm in maximal thickness covered but at the transected area, by the capsule. It shows paramedially a bulging grayish nodule of 3,5 cm in diameter, situated at 1,5 cm from the transected margin. Cut surface is grayish and fibrillary with negative resection margins (Figs. 3–6).

Five months later, during a clinic visit, the patient was without complaint; however it was noted that hypertension had recurred. Repeat CT confirmed 2 new liver lesions – one in segment VI and a second in the papillary process of the caudate. Staging revealed no other disease and these were treated by percutaneous radiofrequency ablation (RFA). Follow-up CT confirmed complete ablation, and hypertension again improved, though briefly. Three months post-RFA, hypertension progressed, and required escalation of her antihypertensive medications. Repeat imaging revealed fairly extensive, multifocal intrahepatic recurrence, but no extrahepatic disease (Fig. 7).

Absent effective chemotherapy, radioembolization was elected, which again controlled the hypertension, but only for a few months. Liver lesions progressed, and the family wished to attempt salvage