CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 78 (2021) 336–339

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Bilateral adrenal hemorrhage after colectomy for perforated diverticulitis: A case report

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ARTICLE INFO

Article history:
Received 6 April 2020
Received in revised form 7 December 2020
Accepted 11 December 2020
Available online 17 December 2020

Keywords:
Adrenal crisis
Adrenal hemorrhage
Colectomy
Perforated diverticulitis

ABSTRACT

INTRODUCTION: Bilateral adrenal hemorrhage can lead to acute adrenal insufficiency. This is a rare complication in the post-operative setting, and we present a case in which it developed after a colectomy for perforated diverticulitis.
PRESENTATION OF CASE: The patient is a 65-year-old female who presented with abdominal pain, nausea, emesis, and hematochezia, and CT scan showing sigmoid diverticulitis with peri-sigmoid abscess. After a failure of non-operative treatment, she underwent Hartmann's resection, and her post-operative course was complicated by refractory tachycardia, hypotension, hyponatremia, and nausea/vomiting. Bleeding, hypovolemia, and sepsis were ruled out. A CT scan showed enlarged poorly defined adrenals bilaterally, suggestive of bilateral adrenal hemorrhage. Serum cortisol level was low and diagnostic of acute adrenal insufficiency. With intravenous steroid therapy (hydrocortisone), her vital signs, laboratory abnormalities, and diet intolerance all resolved. She was discharged on oral prednisone and continued long term.
DISCUSSION: Bilateral adrenal hemorrhage is rare post-operatively and can lead to adrenal insufficiency. 15% of patients who die in shock have bilateral adrenal hemorrhage on autopsy, indicating the necessity of timely diagnosis and treatment of this condition. Corticosteroid therapy is the mainstay of treatment.
CONCLUSION: This case study illustrates that post-operative delay of progression or worsening of condition, with no alternative explanation, can be due to acute adrenal insufficiency resulting from bilateral adrenal hemorrhage, and timely diagnosis and treatment of this condition is paramount for a favorable outcome.

1. Introduction

Bilateral adrenal hemorrhage is a rare phenomenon, reported to have several causes, including sepsis, coagulopathy, prothrombotic state, heparin-induced thrombocytopenia (HIT), and rarely exogenous steroid use. The condition usually presents as acute adrenal insufficiency with abdominal pain, fatigue, hypotension, delirium, hyperkalemia, and hyponatremia. When these symptoms present in a critically ill patient with other comorbidities, a correct diagnosis can be difficult to ascertain. There is often a delay in diagnosis since hypotension is usually attributed to hypovolemia or sepsis.

Early signs and symptoms of acute adrenal insufficiency include abdominal pain, fever, altered mental status, nausea/vomiting, and electrolyte abnormalities [1]. Untreated, it can progress to refractory hypotension and shock. Mortality in acute adrenal insufficiency due to bilateral adrenal hemorrhage is estimated at 15%, though it can be higher if the condition is untreated or caused by certain conditions such as HIT [2].

In this report, we review an unusual case of bilateral adrenal hemorrhage with associated adrenal insufficiency that developed after a colectomy for perforated diverticulitis. This case is presented in accordance with the SCARE criteria [3].

2. Presentation of case

Our patient is a 65-year-old African American woman with a BMI of 26.5, daily smoker with a history of peptic ulcer disease, treated with omeprazole and surgical history of hysterectomy. She presented to the Emergency Department with complaints of worsening left sided abdominal pain, nausea, emesis and hematochezia that had started two weeks earlier. She denied fevers or chills but reported anorexia and a 10-pound weight loss since the onset of her pain. On physical examination, she was afebrile with focalized peritoneal signs in the left lower quadrant without generalized peritonitis. Laboratory data revealed an elevated white blood count (WBC) of 15.3, potassium of 3.0 mmol/L and albumin of 2.3 g/dL.

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https://doi.org/10.1016/j.ijscr.2020.12.028

Table 1
Post operative period.

	HR (avg)	BP (avg)	WBC	Hb/Hct	Na	K	Cl	BUN	Cr	Bowel Function	Steroid administration
Pre-op	68	110/70	15.3	14/43.4	137	3.4	102	22	0.4	No	
POD #1	62	96/56	18	12.2/37.2	140	3.9	104	3	0.5	No	
POD #2	59	107/58	12.7	11.2/32.4	139	3.3	107	4	0.4	No	
POD #3	59	110/56	13.4	11/33.1	137	3.9	105	5	0.4	No	
POD #4	83	111/72	10.3	10.3/31.8	138	4.3	108	4	0.5	No	
POD #5	52	118/62	9.8	11.1/33.4	139	3.7	105	3	0.5	No	
POD #6	79	117/74	*	*	*	*	*	*	*	No	
POD #7	68	151/75	14	10.8/32.7	132	3.4	95	3	0.4	No	
POD #8	102	131/83	18.2	11.7/35.8	134	3.5	95	4	4	No	
POD #9	107	94/63	15	10.4/30.1	133	3.5	95	6	0.4	No	Hydrocortisone 100 mg q8h IV
POD #10	91	95/60	16.3	*	*	*	*	*	*	Yes	Hydrocortisone 100 mg q8h IV
POD #11	63	100/62	*	*	*	*	*	*	*	Yes	Hydrocortisone 100 mg q8h IV
POD #12	65	129/62	10.9	9/26.9	140	4.7	107	16	0.4	Yes	Hydrocortisone 100 mg q8h IV → 50 mg q12 h IV
POD#13	65	148/84	*	*	*	*	*	*	*	Yes	Hydrocortisone 50 mg q12 h IV → 25 mg q12 h IV
POD#14	62	142/73	10.4	10.7/32.9	141	3.2	105	9	0.4	Yes	Hydrocortisone 25 mg q12 h IV → prednisone 5 mg BID PO

*No labs obtained.

Her complete blood count (CBC) and basic metabolic profile were otherwise normal.

Computed Tomography (CT) of the abdomen and pelvis showed sigmoid diverticulitis with a 3.8 × 5.5 cm gas containing pericolonic abscess lateral to the proximal sigmoid colon.

Multidisciplinary management was initiated as the patient was treated with intravenous levofloxacin 500 mg and metronidazole and a 10 fr drain was placed transabdominally by interventional radiology. Twenty ccs of purulent fluid was drained. Microbial cultures from this fluid grew streptococcus viridans, gram negative rods, and proteus species.

The patient continued to have marked abdominal pain with persistent leukocytosis despite drain placement. She was therefore, on hospital day 3, taken to the operating room for an open Hartmann's operation. Final pathology demonstrated perforated diverticulitis with serositis, abscess formation, fibrous adhesions, fat necrosis and granulation tissue, negative for malignancy. Peri-operative antibiotics included IV cefepime 2 mg every 12 h (continued throughout admission), and IV metronidazole 500 mg every 8 h (from two days pre-op through post-operative day (POD) #14), as per discussion with the infectious disease (ID) service.

On POD#1, the patient had mild hypotension and tachycardia despite fluid resuscitation that initially resolved after the epidural infusion was temporarily suspended. Clear liquid diet was started on POD#1. She had minimal intake and no evidence of bowel function. The epidural analgesic catheter was removed on POD#4. Total Parenteral Nutrition was started on POD #5 due to suboptimal oral intake and ileus. On POD#7 the patient had continuous hyponatremia, ranging from to 132–134 mmol/L, potassium of 3.4–3.5 mol/L and hypochloremia of 95 mmol/L (Table 1). On POD #8 CT scan of abdomen and pelvis with PO and IV contrast was obtained due to new onset of emesis and intermittent periods of hypotension and slight sinus tachycardia. It showed new bilateral adrenal enlargement and decreased definition consistent with bilateral adrenal gland hemorrhage/infarction (Fig. 1). A random serum cortisol level was abnormally low at 2.6 ug/dL (normal 10–20 u g/dL). There was no evidence of DVT or PE on lower extremity duplex or chest CTA, and serum cardiac troponin level was undetectable. A transthoracic echocardiogram showed normal ventricular systolic function, EF 60–65%, no valvular disease, and grade 1 diastolic dysfunction. No further cardiac work-up was recommended. Endocrinology was consulted due to the low cortisol level, and a stress dose of hydrocortisone sodium succinate 100 mg was started and given every 8 h. The patient's clinical status improved within the next 24 h. Her blood pressure and heart rate stabilized, and there was return of bowel function. The patient tolerated her diet and was discharged to a rehabilitation facility on POD#14 without TPN or antibiotics. Steroids were tapered down to 5 mg of prednisone twice a day for 2 weeks and 5 mg daily thereafter. The patient was seen in clinic one month after discharge. Her follow up was delayed due to transportation issues. After four months of continued steroid dependence and no signs of adrenal recovery, she was continued on hydrocortisone (20 mg in the morning and 10 mg at night) by endocrinology.

3. Discussion

Bilateral adrenal hemorrhage with associated insufficiency is very rare. Unselected autopsy studies have shown that the prevalence of adrenal hemorrhage ranges from 0.14% to 1.8% [4]. However, 15.4% of patients who died of shock were found to have bilateral adrenal hemorrhage [4]. This condition is most often associated with post-operative patients (especially within 2 weeks after surgery), pediatric patients with meningococcemia (Waterhouse-Frederickson syndrome), thromboembolic events and coagulopathies [2,5].

Different pathogenic mechanisms have been described for this pathology. In the context of sepsis, recent data have shown that meningococcus and other bacteria release endotoxins which enter the sinusoidal spaces of the adrenal gland and stimulate proinflammatory pathways involving IL-1, IL-6 and TNF-α. This process activates fibrinolysis and leads to disseminated intravascular coagulation and hemorrhage [6].

The pathophysiology of spontaneous non traumatic adrenal hemorrhage is thought to be related to the vascular anatomy of the adrenals. The adrenal gland has a rich arterial supply in comparison to the venous drainage, which drains into the medullary sinusoids by a few venules and a single vein susceptible to outflow obstruction [7,8].

Pathological examination of cadavers has shown venous thrombosis in the adrenal glands of patients with bilateral adrenal hemorrhage. It is theorized that this is due to an exaggerated release of catecholamines, thrombin, fibrin and endotoxins [9]. This results

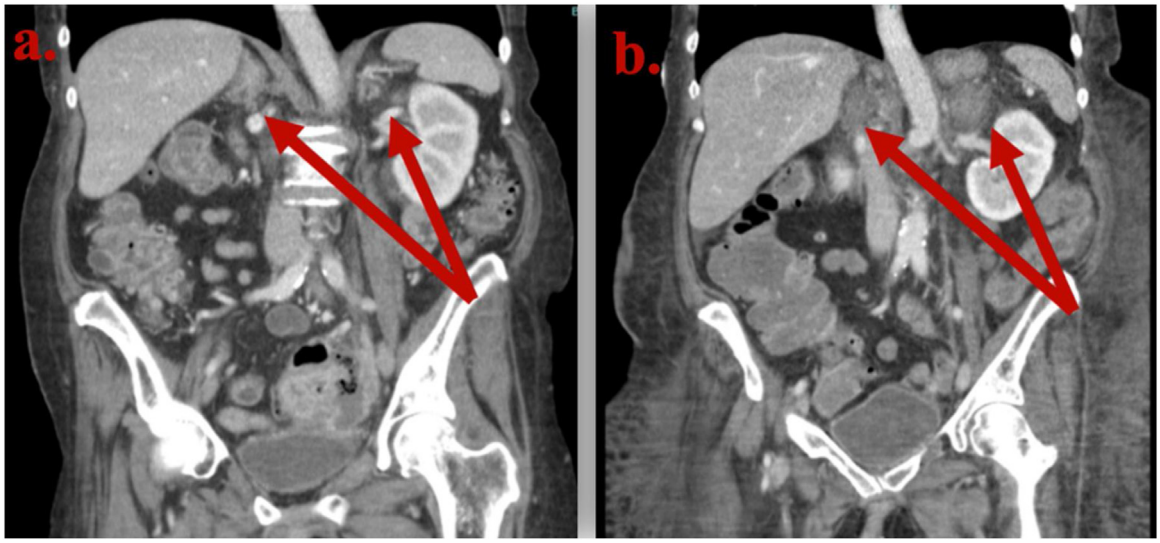

Fig. 1. a. Preoperative CT scan with diagnosis of perforated diverticulitis. (Normal Adrenal Glands). b. Post-operative CT scan with diagnosis of Bilateral Adrenal gland hemorrhage.

in a rise in pressure in the adrenal vasculature, which leads to hemorrhage with associated damage to the adrenal medulla [10].

Previous case reports include cases of adrenal insufficiency due to bilateral adrenal hemorrhage after partial colectomy for malignancy [11,12], and after a small bowel anastomotic leak resulting in sepsis [13]. This is the first report of bilateral adrenal hemorrhage after a colectomy for perforated diverticulitis.

The clinical presentation of acute adrenal insufficiency is nonspecific and includes generalized abdominal pain, back pain, nausea, emesis, electrolyte disbalance specifically hyponatremia and hypokalemia. Due to these vague symptoms, adrenal hemorrhage is often diagnosed late.

Diagnosis of adrenal insufficiency is suggested when early morning, 8 AM, cortisol level is below 10 mcg/dL and confirmed if below 3 mcg/dL (normal range 10–20 mcg/dL). If the cortisol level is borderline reduced, diagnosis can also be supported by performance of a cosyntropin stimulation test. While hyponatremia and hyperkalemia are indicative of adrenal insufficiency, normal electrolyte levels do not exclude the diagnosis.

A postoperative period that deviates from the expected course associated with electrolytes abnormalities should prompt the surgeon to consider bilateral adrenal hemorrhage with associated insufficiency. Initial investigation may include a cortisol level test. Imaging is not part of the algorithm of diagnosing adrenal insufficiency, whether the etiology is related to hemorrhage or not. But if significant adrenal hemorrhage is found incidentally on imaging, it usually appears as a high-attenuating non-enhancing adrenal mass. As the hemorrhage ages, it can become centrally hypoattenuating with peripheral enhancement, and may develop calcifications. The adrenal mass on either side may be round, or it may be adreniform in shape and enlarged [14]. If serial imaging is obtained, the hyperdense adrenal masses will be observed to diminish in size. Acute hemorrhages are usually distinguishable from tumors or hyperplasia on CT given the high attenuation (higher than liver or spleen) [15].

In our case, the patient had persistent hypotension, tachycardia, and ileus. An incidental finding of bilateral adrenal hemorrhage on CT scan lead us to the diagnosis of adrenal insufficiency. We had ruled out other causes of her symptoms, including hemorrhage, sepsis, and hypovolemia.

Once confirmed the management is non operative with supportive measures and evaluation/support of adrenal function with glucocorticoids therapy. This includes intravenous 100 mg hydrocortisone bolus or intramuscular if no IV access. This bolus should then be followed by 200 mg hydrocortisone over 24 h, either as continuous drip or in divided doses. The patient should also receive at least 1 L isotonic saline upfront, followed by followed by a continuous rate, usually aiming for 4–6 liters in the first 24 h, with downward adjustments for patients with heart failure or renal insufficiency [16]. Management should take place in the intensive care unit with close attention to electrolytes and an endocrinology consultation. In the majority of patients, clinical course is temporary and self-limited but serial CT scans or MRIs are highly recommended in case bleeding persists so angioembolization can provide an alternative to surgical exploration [17]. Some patients may require lifelong steroid replacement [12]. Our patient responded well to 300 mg of hydrocortisone daily and fluid resuscitation. She no longer requires steroid therapy.

4. Conclusion

Postoperative bilateral adrenal hemorrhage with associated adrenal insufficiency is rare, but can be lifethreatening. It is essential to be vigilant, and to have a high level of suspicion for this complication, especially when there is a delay of progression or worsening in condition after a major operation.

The diagnosis is based on biochemical testing, and cross-sectional imaging can show adrenal gland hemorrhage. Treatment with steroid replacement is temporary in the majority of cases, but it is important to have serial close monitoring to rule out chronic adrenal insuficiency.

Declaration of competing interest

The authors declare no conflicts of interest pertinent to this case report.

Funding

No source of founding to be declared.

Ethical approval

No institutional review board is required for publication of a case report at our institution.

Consent

Written consent was obtained from the patient directly.

Author's contribution

Esparza Monzavi CA - Study concept/design, data interpretation, writing-original draft preparation, project administration.

Hamed, A - data interpretation, writing-original draft preparation, data interpretation.

Nordenstam, J - writing-review & editing, data interpretation.

Gantt G Jr.- Study concept/design, writing-review & editing, draft preparation, project administration.

Registration of research studies

Not applicable.

Guarantor

Esparza Monzavi CA. MD.
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Provenance and peer review

Not commissioned, externally peer-reviewed.

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