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Case Report

Acute Uterine Torsion Mimicking Tumor Lysis Syndrome

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ARTICLE INFO

Article history:

Received: 16 March, 2020

Accepted: 1 April, 2020

Published: 14 April, 2020

Keywords:

Uterine torsion

tumour lysis syndrome

ABSTRACT

Background: Non-gravid uterine torsion is rare and mostly diagnosed at surgery. This report describes a patient with a large abdominal mass, critical illness, and tumor lysis syndrome.

Case: A female presented with acute abdominal pain. Physical exam showed a large abdominal mass and abdominal rigidity. Serial laboratory tests showed a drop in hemoglobin level, progressive renal failure and serum uric acid and phosphate concentrations compatible with tumor lysis syndrome. Computed tomography showed a 30 cm mass filling the pelvis and abdomen. At an emergency exploratory laparotomy, a 360 degrees torsion of the uterus was found. An uneventful total hysterectomy was done. After surgery renal function restored.

Conclusion: Uterine torsion is rare and can be potentially life threatening when presenting as tumor lysis syndrome.

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Introduction

Acute uterine torsion is a rare cause of sudden onset severe abdominal pain. Most cases occur during pregnancy and torsion of a nonpregnant uterus is extremely rare. Torsion causes obstruction in the blood supply, leading to necrosis and sometimes even distributive shock. We describe a patient who had massive infarction of an enlarged non-pregnant uterus, which was caused by torsion. The laboratory findings were in accordance with the so-called tumor lysis syndrome, which to our knowledge has not been reported previously in a benign gynecological condition. This case should remind clinicians that uterine torsion is one of the potential diagnoses in patients with fibroid uterus and acute onset of abdominal pain. Further, this case informs clinicians on the potential occurrence of the tumor lysis syndrome in a benign condition.

Case

A 49 years old female with a history of bipolar disorder with psychotic episodes was known with a large fibroid uterus for years. She was treated with lithium, pimozid and trihexyfenidyl for her schizophrenia, and used

lynestrol and paracetamol for bleeding problems and pain of the fibroid uterus. Fear for post-surgery psychosis made her refuse recommended hysterectomy. She was admitted to a referring hospital with acute onset, severe, progressive abdominal pain, nausea and vomiting. Physical examination revealed a large, firm, painful mass in the abdomen, extending from the symphysis to the xiphoid. She had abdominal rigidity and on pelvic exam severe tenderness. Trans abdominal ultrasound showed a large mass, filling the pelvis and full abdomen. Ovaries and uterus could not be distinguished. A computed tomography was performed without contrast medium, since renal failure was present. This confirmed the presence of a 30-cm mass situated on the right side of the abdomen, in tight contact with the right ovary, filling the abdomen. Peritoneal fluid was seen extending beyond the pelvis.

Although hydronephrosis was not seen on computed tomography images the renal dysfunction was assigned to obstruction of the ureters and a diminished circulating volume. She developed tachycardia, tachypnea and consciousness decreased. Admittance to the intensive care unit (ICU) followed, for hemodynamic monitoring and fluid suppletion. Urine production increased slightly. Because of lithium intoxication (lithium level 1.6 mmol/L) and renal failure, the lithium was stopped.

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The pimozid and trihexyfenidyl were replaced by olanzepin. Cephalosporin was started to treat a suspected sepsis, as CRP and leucocyte levels were highly increased and vital parameters deteriorated. The full set of laboratory results is depicted in (Table 1). Despite these interventions renal failure aggravated. Tumor lysis syndrome was suspected because of elevated potassium and uric acid levels in combination with progressive renal failure (Table 1). Also, a decrease in hemoglobin level from 7,4 to 3,8 mmol/L was observed raising the suspicion of a major intra-abdominal hemorrhage. The patient was transferred to our tertiary clinic for exploratory laparotomy and emergency hysterectomy, with embolization of the uterine arteries prior to surgery.

Table 1: Laboratory results on subsequent days.

Element	Day -1	Day 0	Day +1	Reference
Hemoglobin	7.4	3.8	5.1	7,5-10 mmol/L
Platelet count	245	189	-	150-400 x 10 ⁹ /l
White blood cell count	43.7	41.7	33.4	4.5-11.0 x 10 ⁹ /L
C-reactive protein	224	431	295	<10 mg/L
Creatinine	280	292	197	45-90 µmol/l
e-GFR (MDRD)	17	16	23	> 60 ml/min
Urea	21.4	17.7	17.7	2,5-7,5 mmol/l
Uric acid	0.79	-	-	0,20-0,42 mmol/l
Sodium	128	131	142	136-144 mmol/l
Potassium	5.7	5.1	4.2	3,5-5,0 mmol/l
Calcium (non-ionized)	-	2.13	1.04	2,10-2,55 mmol/l
Albumin	-	-	18	35-55 g/L
Phosphate (mmol/l)	1.27	1.52	-	0,9-1,45mmol/l
Lactic acid dehydrogenase	471	643	518	130-250U/l

Table 2: Classification system for tumor lysis syndrome, according to Cairo and Bishop [3].

<i>Laboratory tumor lysis syndrome:</i> abnormality in two or more of the following, occurring within three days before or seven days after chemotherapy.
• uric acid $\geq 0,476$ mmol/l or 25% increase
• potassium ≥ 6 mmol/l or 25% increase
• phosphate $\geq 1,45$ mmol/l or 25% increase
• calcium $\leq 1,75$ mmol/l or 25% decrease
<i>Clinical tumor lysis syndrome:</i> laboratory tumor lysis syndrome plus one or more of the following:
• increased serum creatinine (1.5 times upper limit of normal)
• cardiac arrhythmia or sudden death
• seizure

At presentation in our clinic (day 0 in table 1) the patient was extremely painful, but responsive. She was admitted to our ICU with a blood pressure of 90/40mmHg, pulse rate 110 bpm and core temperature was 38.2°C. Urine production was less than 10 cc an hour. Laboratory tests in our clinic strengthened the suspicion of tumor lysis syndrome, with

renal failure, elevated potassium, uric acid and now also increased phosphate levels (Table 1).

Because of the decline in clinical condition, laboratory tests and presumed bleeding, we proceeded with uterine artery embolization and hysterectomy. Surgical exploration showed an enlarged, purple colored uterus with a leiomyoma (size 30 cm), and purple colored adnexa. The internal genitals had undergone torsion, 360 degrees in rotation. An uneventful, supravaginal hysterectomy and bilateral salpingo-ovariectomy was performed. Histopathological exam confirmed a leiomyoma of 30 cm and the presence of infarction, congestion and necrosis as well as extensive hemorrhage within the uterus and leiomyoma. The ovaries and tubes were normal and there were no signs of malignancy. Laboratory abnormalities improved immediately after surgery (Table 1). In the postoperative period the patient developed psychosis and unexplained polyneuropathy.

Discussion

This case describes a woman with massive infarction of an enlarged torted fibroid uterus, resulting in tumor lysis syndrome and requiring emergency treatment. To our knowledge this is the first case described with tumor lysis syndrome in a benign gynecological condition. Tumor lysis syndrome refers to the constellation of metabolic disturbances that may follow the initiation of cancer treatment. After cell lysis, phosphate, potassium and nucleic acid are released into the circulation. Catabolisation of nucleic acid results in hyperuricemia leading to the precipitation of crystals and depositions in the collecting renal tubules and finally to renal impairment and oliguria. The cell lysis also leads to hyperkalemia, hyperphosphatemia and secondary hypocalcaemia. Generally, hyperkalemia is the first electrolyte imbalance to occur, followed by hyperphosphatemia, similar to our case. Serum calcium binds to the elevated phosphorus within the bloodstream, causing hypocalcaemia. The electrolyte abnormalities result in gastrointestinal complaints such as nausea, vomiting and abdominal cramping [1, 2].

In 2004, Cairo and Bishop defined a classification system for tumor lysis syndrome, (see Table 2) [3]. Tumor lysis syndrome usually occurs in patients with bulky, rapidly proliferating, treatment-responsive tumors and is classically associated with hematologic malignancies following recent chemotherapeutic treatment. However, it can also develop in patients with solid tumors and in the absence of recent chemotherapy. It is a potentially lethal complication. Risk factors for lethality include high lactate dehydrogenase levels ($> 2 \times$ reference value), elevated urea, pre-existing impaired renal function and oliguria. The management of tumor lysis syndrome includes avoidance of nephrotoxic drugs, aggressive hydration and forced diuresis with diuretics. The latter two are fundamental to enhance the excretion of uric acid and phosphate, restore intravascular volume with increased renal blood flow and glomerular filtration. Potassium binders, phosphate binders and allopurinol or rasburicase should be administered [1, 2, 3]. In our case the patient received extensive fluid suppletion and post-surgery forced diuresis. Nephrotoxic drugs were avoided. No binding medicines were administered. Lithium is known to cause renal failure when prescribed in high dosage for long periods. Our patient was treated with lithium over ten years. Although we have no information on preceding renal function

tests, it can be that the sub-clinical renal failure contributed to the deterioration in renal function.

Uterine torsion is rare. It usually occurs in pregnancy. Only a few cases of a non-gravid uterine torsion have been described [4-8]. A large leiomyoma or ovarian mass is the most common predisposing factor. Other predisposing factors are pelvic adhesions or uterine malformations. Torsion always occurs along the transition of corpus and cervix uteri and rotation is mostly clockwise. Rotation around the vertical axis of more than 45° defines a pathologic uterine torsion, but torsions up to 720° have been described. Generally, the round and broad ligaments prevent the uterus from torsion. In case of a torsion these ligaments even as the cervix are extremely stretched [4, 5, 7]. Symptoms accompanying torsion of the uterus are non-specific and varying from mild to severe pain, shock, vaginal bleeding and gastro-intestinal symptoms. Findings at physical examination can be uterine tenderness, a large uterine mass in case of a fibroid uterus, cranial extrusion of the cervix and a twisted vaginal canal. Imaging techniques can be helpful diagnosing uterine torsion, but the definite diagnosis is nearly always made intraoperatively [7, 8]. The treatment of choice is surgery, in particular when there is suspicion for infarction or necrosis. Untwisting the uterus manually should be avoided since embolic complications have been described [4, 7].

In our case the patient reported abdominal pain and complaints of nausea. The medical history of a fibroid uterus, the physical examination, the decrease in hemoglobin level and the computed tomography were interpreted as major hemorrhage in an enlarged fibroid uterus. During surgery there was no bleeding and in retrospect it is more likely that the decrease in hemoglobin level was caused by the uterine torsion itself. In uterine torsion, first venous drainage impairs, with congestion of blood in the uterus. Second, the arterial supply is hampered causing cell death. In a large size torsed uterus, like in our case, a shift in blood flow from circulation to the third space of the uterus can be substantial, resulting in a significant drop in hemoglobin level without actual bleeding. Uterine artery embolization was performed before surgery since a technically difficult procedure with preceding hemorrhage was expected. Only upon surgery the definite diagnosis of uterine torsion was made. The subsequent hysterectomy was uneventful. It is not known whether the foregoing embolization affected this outcome.

The pre-operative laboratory abnormalities (hyperuricemia and hyperphosphatemia) and acute kidney failure were interpreted as pointing to the clinical tumor lysis syndrome. However, in our case there was no malignancy. A possible explanation for the tumor lysis syndrome in this patient is the major amount of necrosis and cell lysis in the large torsed fibroid uterus resulting in the release of intracellular components

into the bloodstream as is seen in the tumor lysis syndrome in malignancies. Post-operative our patient recovered well without any signs of permanent renal damage; she did however develop a psychosis. Whether this is caused by the combination of the hospital admission, the poor clinical condition, the laparotomy or the temporary interruption of lithium is not clear. Unfortunately, the psychosis was the one thing she was most afraid of and the reason why she refused the surgery since years before.

Teaching points

- In case of sudden onset severe abdominal pain in women with an enlarged fibroid uterus, the diagnosis uterine torsion should be considered.
- Tumor lysis syndrome, a potentially life-threatening illness, can be the presenting symptom of uterine torsion.

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