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Laparoscopic hysterectomy as optimal approach for 5400 grams uterus with associated polycythemia: A case report

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Abstract

BACKGROUND

Uterine myomas are the most common benign gynecological soft tissue tumors in women but polycythemia associated with these tumors is rare. Polycythemia is significantly associated with risk of venous thromboembolism (VTE), which is further increased in case of a large pelvic mass and obesity. Here we report the surgical challenges in the case of an obese patient with a giant fibromatous uterus and associated polycythemia.

CASE SUMMARY

A 42-year-old obese woman was referred to our department for treatment for a huge fibromatous uterus. She suffered of pelvic pressure and abdominal distension and reported severe dyspnea from several months. Laboratory analyses including hemoglobin (Hb) 19.2 g/dL and hematocrit (Hct) 59.7% were indicative of polycythemia. Arterial blood gas analysis showed arterial oxygen pressure (pO₂) of 81.5 mmHg. Abdominal computed tomography confirmed a giant fibromatous uterus measuring 28.2 cm × 17 cm × 25 cm. To reduce the thromboembolic risk, the patient underwent low molecular weight heparin, phlebotomy twice before surgery, and we opted for a laparoscopic hysterectomy. The uterus weighed 5400 g and the histology confirmed a diagnosis of leiomyoma. We did not find increased erythropoietin levels in the supernatant of the myomatous uterine tissue. There were no complications. On postoperative day 1, the Hb and the Hct levels normalized to 13.3 g/dL and 41.7%, respectively. Arterial blood gas analysis after surgery showed a pO₂ of 144.7 mmHg. These results suggested the diagnosis of myomatous erythrocytosis syndrome. The patient was discharged on the second postoperative day in very good condition with no symptoms.

CONCLUSION

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We believe this is the first reported laparoscopic hysterectomy in a patient with a giant uterus, polycythemia and obesity. Multiple VTE risk factors warranted a laparoscopic approach.

Key words: Hysterectomy; Laparoscopy; Myoma; Myomatous erythrocytosis syndrome; Polycythemia, huge uterus; Total laparoscopic hysterectomy; Case report

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Core tip: Uterine myoma are rarely associated with polycythemia. Here we report the challenging case of an obese patient with a giant fibromatous uterus and associated polycythemia; these factors increased significantly the risk of venous thromboembolism. To reduce such risk, we opted for a laparoscopic hysterectomy, which was preceded by phlebotomy. Laparoscopy was successfully completed without complications. The uterus weighed 5400 g. After surgery the patient's hematological profile normalized. The final diagnosis was myomatous erythrocytosis syndrome (MES). To our knowledge, this is the first case of laparoscopic hysterectomy in an obese patient with a very large uterus and polycythemia due to MES.

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INTRODUCTION

Uterine myomas are the most common benign gynecological soft tissue tumors in women but polycythemia associated with these tumors is rare^[1-3]. In 1953 Thomson and Marson described the first patients with polycythemia linked to uterine myomas, in which tumor excision resulted in the cure of this hematological disorder^[4]. In 1957 Fleming *et al*^[5] defined this condition as myomatous erythrocytosis syndrome (MES) when associated with polycythemia, a myomatous uterus, and a return to a normal hematological profile following hysterectomy or myomectomy.

Typically, secondary polycythemia is related to defective lung function, congenital heart disorder and tumors^[6]. In MES it is believed that polycythemia is strictly linked to the excessive production of erythropoietin (EPO) by the tumors^[7], but oxygenation disorders in the presence of a large abdomino-pelvic mass may also be the cause^[8]. Complications of polycythemia include a significantly increased risk of arterial and venous thromboembolism (VTE)^[9] and pose a problem in the choice of surgical approach. In fact, in this syndrome the polycythemia is typically resolved with the removal of the tumor and therefore the choice of surgical approach is crucial in patient's management. It is also known that the surgery and the time of bed-rest and convalescence are associated with increased rates of VTE^[10]. Here we discuss the case of an obese patient with a giant fibromatous uterus and polycythemia that raised challenges about the choice of surgical approach. In fact, it is known that both obesity and a large pelvic mass are common risk factors for venous thrombosis. The invasive venography or contrast-enhanced computed tomography have exactly showed that large uterine fibroids can constrict pelvic veins^[11], and it is known that obesity further increases the thrombotic risk in all types of surgery^[12].

To our knowledge this is the first described report of a laparoscopic hysterectomy performed in a patient with a giant uterus (5400 g), polycythemia and obesity.

CASE PRESENTATION

Chief complaints

A 42-year-old woman was referred to our department for treatment for a huge fibromatous uterus.

History of present illness

Her symptoms included pelvic pressure and abdominal distension and for several months' severe dyspnea.

Past medical history

Her medical history included a thyroidectomy for a thyroid cancer under treatment with L-thyroxine.

Personal and family history

The patient was gravida 0, para 0.

Physical examination

Her weight was 85 kg and height 156 cm (body mass index 35). At physical examination her abdomen appeared filled by a pelvic mass arriving at the xiphoid process.

Laboratory examinations

Laboratory analysis produced the following results: red blood cell count, $7.04 \times 10^6/\text{mm}^3$; hemoglobin (Hb), 19.2 g/dL; hematocrit (Hct), 59.7%; white blood cell count, $9400/\text{mm}^3$; platelets $325/\text{mm}^3$; iron 47 mcg/dL; ferritin 14 mcg/L; transferrin 310 mg/dL; C-reactive protein 0.5 mg/dL (range ≤ 0.10 mg/dL); serum levels of EPO 45 mIU/mL (normal range: 0-29.0 mU/mL); Ca-125: 11.9 U/mL; Ca 19-9: 26 U/mL; Ca 15.3: 41 U/mL; and He4: 34.4 pmol/L. These results were in line with the diagnosis of polycythemia^[13,14].

The arterial blood gas analysis showed arterial oxygen pressure (pO₂) of 81.5 mmHg (pCO₂ 38.4 mmHg; Oxygen saturation, sO₂ 96.2%; Hct 61%; and total Hb 20.6 g/dL). The preoperative cytology of the endometrium was normal.

Imaging examinations

Preoperative abdominal and transvaginal ultrasound showed a huge fibromatous uterus. The abdominal CT scan (Figure 1 and Figure 2) confirmed a giant fibromatous uterus with important vascularization and inhomogeneous contrast enhancement, with clear limits and regular margins measuring 28.2 cm × 17 cm × 25 cm. The mass displaced the ileal loops upward and to the left.

FINAL DIAGNOSIS

These results suggested the diagnosis of polycythemia associated with a giant fibromatous uterus (MES).

TREATMENT

To reduce the thromboembolic risk in the perioperative period, the patient underwent treatment with low weight molecular heparin (enoxaparin 4000 UI/die), phlebotomy two times before surgery, and to further reduce this fearsome complication we opted for a laparoscopic hysterectomy approach (total laparoscopic hysterectomy, TLH). After the phlebotomies, the day of surgery, the patient's preoperative Hb and Hct values were reduced to 16.8 g/dL and 51.2%, respectively.

We performed the surgery with the patient under general anesthesia in the lithotomy position. We placed a nasogastric tube to reduce the risk of stomach distention and perforation. For better uterus manipulation, we utilized the ClearView® Uterine Manipulator-7 cm (Ethicon Endo-Surgery, Cincinnati, United States). A 12-mm trocar was placed exactly under the xiphoid process and a pneumoperitoneum of 10-14 mmHg was generated in a neutral position and kept for the whole duration of the surgery. Intra-abdominal view was obtained with a 10 mm, 0° telescope (Karl Storz, Tuttlingen, Germany). We placed two 5-mm trocars in the hypochondrium lateral to the rectus abdominis muscle, a third 12-mm trocar at the umbilicus and another 5 mm trocar in the suprapubic position. TLH was then carried out in a consolidated technique reported in detail in our previous manuscripts^[15,16]. To perform all the procedures, we used the LigaSure™ (Medtronic; AutoSuture Co., Norwalk, CT) and the BiClamp® LAP forceps (ERBE GmbH, Tübingen, Germany). Once completed the extrafascial hysterectomy, we extracted the uterus through a very low transverse laparotomic incision of about 5 cm, utilizing a wound protector/retractor (Wound Edge Protector - 3M™ Steri-Drape™ 1073, Diegem, Belgium) to safeguard the cut site, and morcellated it using a cold blade scalpel outside to avoid spillage.

At the conclusion of surgery, following the suture of the accessory laparotomy, we



Figure 1 Preoperative computed tomography imaging. A, B: Coronal (A) and sagittal (B) scans of the huge myomatous uterus occupying the entire abdomen: The orange lines show its major diameters.

laparoscopically examine the abdominal cavity and washed it carefully to remove any possible residue. There were no complications; the operative time was approximately 220 min. Intraoperative blood loss was ≤ 10 ml. The arterial blood gas analysis after surgery showed a pO_2 of 144.7 mmHg (pCO_2 40.4 mmHg; sO_2 98.8; Hct 45%; tHb 15.4 g/dL).

OUTCOME AND FOLLOW-UP

The weight of the uterus was 5400 g. The histological examination confirmed the diagnosis of leiomyoma. In order to determine whether the uterine leiomyoma was producing EPO, 200 mg frozen samples of the myoma tissue were blended with 500 mL of lysis buffer including the tissue protein extraction reagent and protease inhibitor mixture (Thermo Scientific). The supernatant was gathered by centrifugation at $10000 \times g$ for 5 min, and the EPO levels were analyzed by a human EPO enzyme-linked immunosorbent assay (ELISA) (DRG Instruments GmbH, Marburg, Germany). A same size sample of myoma tissue from a patient without polycythemia was used as a control. The EPO level in the myomatous tissue of our patient was 1.5 mIU/mg, similar to the level found in the control sample (1 mIU/mg).

The patient's postoperative course was wonderful. On postoperative day 1, the Hb level had normalized to 13.3 g/dL, and the Hct level had fallen to 41.7%. The patient left the hospital on the second postoperative day in very good general status and with no symptoms.

Repeated blood analyses after 1 wk (Hb 12.6 g/dL, Hct 38.7%) and 4 mo (Hb 12.8 g/dL, Hct 40.6%), confirmed the normalization of the patient's hematological profile, so we were able to report the case in question as a MES.

DISCUSSION

Hysterectomy for uterine fibromatosis is the most common procedure in gynecologic surgery^[17]. Surgeons are increasingly employing laparoscopy for hysterectomy^[18]. However, the role of laparoscopy in the presence of giant uteri is still debated. Siedhoff *et al*^[19] carried out a decision analysis comparing laparoscopic hysterectomy to abdominal hysterectomy for the treatment of the large uteri with supposed benign myomas, examining mortality, surgical complications, and quality of life. They demonstrated a decreased overall mortality associated with laparoscopic hysterectomy and morcellation compared to laparotomic approach. In particular, the laparoscopic group had lesser incidence of VTE (690 vs 840 per 100000) associated with reduced bedrest compared to the abdominal group^[19]. This evidence finds its emphasis in clinical conditions such as large uterine volume and obesity that predispose to VTE, and becomes even more important in rare conditions such as polycythemia associated with uterine fibromatosis, that further increase the risk. The



Figure 2 Preoperative computed tomography imaging. Axial scan of the huge myomatous uterus showing its diameter (orange line).

clinical case described here is the first relative to laparoscopic hysterectomy in a patient with MES and a giant fibromatous uterus (5400 g). The polycythemia associated with this clinical case and the possibility of an associated VTE strongly suggests the need for a careful choice of the hysterectomy procedure. There is no doubt that laparoscopic hysterectomy has certain advantages over abdominal hysterectomy in terms of less bed rest and convalescence time and should be considered the option of choice, especially in conditions predisposing to thromboembolism^[20]. The problem of the volume of the uterus still influences the choice of the most adequate surgical procedure, but this clinical case demonstrates once again that laparoscopic hysterectomy is feasible and free of side effects even in case of giant uterus^[15,16].

Notably, this patient's polycythemia did not depend on uterine EPO production but was probably due to the excessive number of months in which the patient suffered dyspnea with a physiological obstacle to the oxygenation of the blood due to the large volume of the uterus.

CONCLUSION

This study highlights the need for careful screening so that women suffering from uterine fibromatosis are treated before this results in excessive uterine volume with potentially very severe related symptoms. Laparoscopic hysterectomy should be considered as the optimal surgical approach to reduce the risk of VTE, even in cases of very large uteri.

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