Clinical considerations and sonographic findings of a large nonpedunculated primary cervical leiomyoma complicated by heavy vaginal haemorrhage: A case report and review of the literature

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Summary

Leiomyomas of the uterine cervix are uncommon. Cervical leiomyomas in non-pregnant women rarely are of clinical significance and their complications include pressure effects on the bladder or urethra, degenerative phenomena and menorrhagia. We present a case of 46-year-old female, gravida 2, para 1 who was presented in the gynaecological emergency room with the chief complaint of profound vaginal bleeding over the previous three weeks with recent passage of clots. Pregnancy test was negative. She suffered from profound orthostatic hypotension and tachycardia. On examination, her abdomen was soft, non-tender, and without rebound, guarding or palpable masses. Bimanual examination was notable for the presence of a large firm mass fixed to the uterine cervix. The cervix was 8 cm dilated and the body of the uterus was felt separate from the mass. The sonographic findings were consistent with a large cervical leiomyoma. Subtotal hysterectomy with salpingo-oophorectomy was performed and the patient’s postoperative course was uneventful. In conclusion, in the present case the cause of the heavy vaginal bleeding was a primary cervical fibroid as the uterus attempted to abort it.

Key words: Cervical leiomyoma; Cervical fibroid; Vaginal bleeding; Complications; Ultrasonography.

Introduction

Leiomyomas (fibroids or myomas) are benign tumors that arise from smooth muscle cells and represent the most common pelvic tumors of the female genital tract [1]. The vast majority of leiomyomas usually occur in the uterine body. However, they also can occur on the uterine cervix, the vagina, the vulva, the fallopian tubes, the ovaries and the uterosacral ligaments [1-4].

Uterine leiomyomas are clinically apparent in about 25% of women, and with newer imaging techniques, the true clinical prevalence may be higher [5]. Uterine fibroids are estimated to produce symptoms at a rate between 20% and 50% [6-8]. Symptoms attributable to uterine leiomyomas can generally be classified in three distinct categories: (i) abnormal uterine bleeding, (ii) pelvic pressure and pain, and (iii) reproductive dysfunction [5]. In addition, haemoperitoneum can result from perforation of a cystic degenerated subserosal leiomyoma [9]. Cervical leiomyomas usually present urinary problems as they displace the urinary bladder and uterus [10]. Also, these tumors can cause dystocia in pregnant women and can be complicated by the same degenerative phenomena that occur in leiomyomas of the uterine corpus [2] and have the same risk for malignant transformation. Cervical leiomyomas producing menorrhagia are very unusual [10].

The present case highlights the ultrasound appearance and the management of a rare case with a large nonpedunculated primary cervical fibroid in a 46-year-old patient who presented with prominent uterine bleeding and cervical dilation of 8 cm. On admission she suffered from profound orthostatic hypotension, tachycardia and her haemoglobin was 5.8 g/dl. Although in non-pregnant women cervical leiomyomas are usually without clinical significance, this paper presents an additional case of primary cervical leiomyoma associated with heavy vaginal bleeding.

Case Report

A 46-year-old female, gravida 2, para 1, presented to the emergency gynaecological room of the “George Gennimatas” General State Hospital of Athens, Greece with the chief complaint of profound vaginal bleeding over the previous three weeks with recent passage of clots. One year prior to her presentation at our hospital, the patient reported irregularity of her menstrual cycles with periods of heavy menorrhagia. Gynaecological history consisted of regular menstrual cycles every 28 to 30 days since menarche at the age of 14. She also had a history of an induced abortion. Her family and her medical history were unremarkable. She had no known coagulopathy and no history.
of pelvic inflammatory disease. She took no medications or oral contraceptives and had no allergies.

On admission, physical examination disclosed a well-nourished and well-developed female. She was markedly pale. She suffered from profound orthostatic hypotension and tachycardia. Her abdomen was soft, non-tender, and without rebound, guarding or palpable masses. Pelvic examination disclosed normal external genitalia. The vaginal vault contained fresh blood clots. Bimanual examination was notable for the presence of a large firm mass fixed to the uterine cervix. The uterine cervix was 8 cm dilated and the body of the uterus was felt separate from the mass. The clinical differential diagnosis included an aborting pedunculated intracavitary uterine leiomyoma or a cervical leiomyoma.

The electrocardiogram and chest radiography were normal. Complete blood cell count at this time showed haemoglobin 5.8 g/dl, haematocrit 18.4%, white blood count 8.8x10³ cells/mm³ and platelet count 221,000/mm³. The values for urea nitrogen, serum electrolytes, prothrombin time and activated partial thromboplastin time were normal. The pregnancy test was negative. Urinalysis was notable for a large amount of blood and with no bacteria noted. Vaginal sonography demonstrated a large (maximum diameter 9.1 cm) solid mass of inhomogeneous texture in the region of the cervix (Figure 1a). The actual body of the uterus as well as the uterine adnexa could not be disclosed because of the size of the mass. The endometrial thickness was 13.3 mm (Figure 1b). Transabdominal sonography (the urinary bladder was empty) revealed a large round, solid, heterogeneous and well circumscribed mass in the uterine cervix (Figure 2). This structure was consistent with a large cervical leiomyoma. The body of the uterus was normal and both adnexa appeared normal.

The patient was admitted to the hospital for surgical management and was transfused with two units of packed red blood cells prior to the operation. In the operating room she was placed in the dorsal lithotomy position and under general anaesthesia her vaginal and vulvar preparation was done with aqueous betadine solution. She was fully draped in a sterile fashion. An attempted transcervical resection of the leiomyoma failed. The patient was placed in the usual surgical position and the abdomen was entered in layers through a Pfannenstiel incision. The patient was placed in a moderate Trendelenburg position and the intestines were packed up into the upper abdominal cavity using abdominal packs soaked in Ringer’s lactate solution. The uterine body had a regular size and the cervix was found bulky. Both adnexa appeared normal. Bilateral clamping, cutting and ligation of the round and infundibulopelvic ligaments were done respectively. The vesicouterine and rectovaginal folds were cut and the bladder was mobilized. The uterine vessels were easily clamped, cut and ligated. A vertical incision was made on the anterior cervical wall using a monopolar electrode of the electrosurgical unit arranged in the cutting wave-form; the cervical cavity was entered easily (Figure 3). A large submucosal firm and fixed tumor in the cervix was found. The tumor was cut from the cervical wall using the monopolar electrode in conjunction with counter-traction with a single-tooth tenaculum. After that, subtotal abdominal hysterectomy with bilateral salpingo-oophorectomy was performed. The cervical cuff was closed and the pelvis reperitoneализed. The blood loss during surgery was unremarkable. The patient was transfused with one unit of packed red cells and one unit of fresh frozen plasma during the operation and with the same units of packed red cells and fresh frozen plasma on the first postoperative day, respectively. Her immediate postoperative condition was satisfactory and she was discharged on the seventh postoperative day.

On follow-up four weeks later the patient was very well.

Histology reported a benign cervical leiomyoma, measuring 9.5 x 8 x 4 cm. The surface, in an area with a diameter up to 3 cm, was covered by squamous mucosa. The body of the uterus measured 5 x 4.7 x 3 cm and the endometrial cavity 4 x 3 cm. The thickness of the myometrium was 1.5 cm. Five additional leiomyomas were found in the uterine corpus with a maximum diameter between 0.4 and 1.8 cm. Also, foci of adenomyosis were noted. The endometrial mucosa was found to be at the secretory phase. Both fallopian tubes and ovaries were microscopically normal.
prominent uterine bleeding and haemoglobin of 5.8 g/dl. The absence of any underlying disease, the negative pregnancy test and the pelvic examination were suggestive of an aborting pedunculated intracavitary uterine leiomyoma or a cervical leiomyoma. With the ultrasonographic examination the presumptive diagnosis of a large cervical fibroid was made. It is interesting to note that transabdominal sonography rather than transvaginal sonography proved to give particularly helpful information concerning the actual location of the mass, which was decisive in the final sonographic diagnosis of the cervical leiomyoma. After surgery the pathology report showed apart from the large cervical fibroid, the presence of five small leiomyomas in the uterine corpus. However, we believe that the cause for the heavy vaginal bleeding was the large cervical fibroid because the uterus tried to abort the cervical fibroid and the cervix was found to be significantly dilated. The cervical leiomyomas in non-pregnant women are rarely of clinical significance. The isolated cases leading to complications are noted because of their infrequency and include pressure effects on the bladder or urethra, and degenerative phenomena such as red, hyaline, myxomatous, mucoid, fatty or cystic degeneration [2, 9, 10]. Cervical leiomyoma associated with menorrhagia as in our patient is very unusual.

In conclusion, this was a case of an unusual presentation of a primary cervical fibroid. Interesting ultrasonographic findings and the surgical technique for a subtotal abdominal hysterectomy for cases with large cervical fibroids were described.

References


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Torsion of a non-gravid leiomyomatous uterus in a patient with myotonic dystrophy complaining of acute urinary retention: anaesthetic management for total abdominal hysterectomy

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Summary

Torsion of a pregnant uterus is rare, but torsion of a non-pregnant uterus is extremely rare. Abdominal pain is the major symptom. Other symptoms include vaginal bleeding, urinary tract symptoms and gastro-intestinal manifestations. We present a case of a 37-year-old white nullipara who presented at the emergency room with acute urinary retention. Medical history revealed that the patient carried the disease of myotonic dystrophy, which was diagnosed two years before. Physical examination revealed a tender, distended bladder, which was easily catheterized, draining 900 ml of clear urine. The abdomen was soft with no muscle guarding or rebound tenderness. A palpable large dense mass occupying the cul-de-sac was found during bimanual examination. Abdominal ultrasound examination revealed a large intramural leiomyoma approximately 10 cm in diameter, in the posterior wall of the uterus, which repelled the bladder. In neurological examination the muscular tone and reflexes were reduced in the lower extremities. Myotonic phenomenon was not found. The patient was thought to suffer from myotonic dystrophy and therefore the possibilities for pulmonary and cardiac complications or malignant hyperthermia had to be kept in mind during the anaesthetic management. The patient underwent an exploratory laparotomy and the uterus was found to have undergone a 60° rotation along the corpus and the cervix uteri transition line. Total abdominal hysterectomy with bilateral salpingo-oophorectomy was performed. The intra- and postoperative course of the patient was uneventful. In conclusion, in this patient the uterine pathology (large leiomyoma) in combination with the disease of myotonic dystrophy seemed to be the predisposing factors for the torsion of the non-pregnant uterus. Also, the anaesthetic implications for total abdominal hysterectomy in myotonic dystrophy are discussed and the international literature is reviewed.

Key words: Uterine torsion; Twisted uterus; Leiomyoma; Acute urinary retention; Myotonic dystrophy; Anaesthetic management; Total abdominal hysterectomy.

Introduction

Myotonic dystrophy is a dominantly inherited disease, with a prevalence of five per 10,000 in Europe and North America and an exceptionally high prevalence in Quebec, Canada [1, 2]. It is genetically transmitted autosomically and is the most common hereditary muscular illness not related to the sex [3]. This disease is characterized by increased muscular irritability and contractility, with decreased power of relaxation, muscle wasting, cataract, premature frontal baldness, gonadal atrophy, hyperostosis cranii, disorders of cardiac conduction and endocrine dysfunction [4]. As the disease progresses, muscle wasting and weakness occur, involving facial muscles, muscles of mastication, sternocleidomastoids, thenar, forearm, quadriceps, and pedal dorsiflexors [4]. Chronic respiratory failure in myotonic dystrophy is a frequent complication that can lead to premature death, but the incidence is uncertain [2].

Torsion of pregnant uterus is rare, but torsion of a non-pregnant uterus is extremely rare and is defined as rotation of more than 45° around the long axis of the uterus [5]. In two-thirds the rotation takes place towards the right. It appears that uterine axial torsion is usually caused by the presence of a pathological or abnormal condition in the uterus or the adjacent structures [5]. Clinical manifestations could be acute, chronic or asymptomatic [6].

We report an extremely rare case, in which acute urinary retention was caused by torsion of a non-pregnant leiomyomatous uterus in a patient suffering from myotonic dystrophy. The patient underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy and her intra- and postoperative course was benign. The anaesthetic implications in myotonic dystrophy are discussed and the international literature is reviewed.
Case Report

A 37-year-old Caucasian white woman, (gravida 0, para 0), was admitted to the “George Gennimatas” General State Hospital of Athens because of difficulty with micturition characterized by acute weakness to pass urine. The first menstrual bleeding occurred at the age of 11. Menstrual cycles were regular, every 28 to 30 days and lasted for five to six days. She had had a laparotomy for uterine myomectomy seven years prior to her admission in our hospital and an operation for bilateral cataracts. For the last five years, she had been treated for a hypothyroid condition with 0.1 mg of levothyroxine sodium daily. There was familiar myotonic dystrophy whose genealogy was confirmed. The patient was aware that she carried the disease, which was diagnosed two years earlier.

Physical examination revealed a tender, distended bladder, which was easily catheterized, draining 900 ml of clear urine. The abdomen was soft with no muscle guarding or rebound tenderness. A palpable large dense mass occupying the cul-de-sac was found during vaginal examination; no uterine tenderness was noted. Her blood pressure, pulse and temperature were normal. White blood count on admission was 12,300 cells/mm³ with 85% polymorphonuclears and haemoglobin at 14 g/dl. Abdominal ultrasonography revealed a large intramural myoma approximately 10 cm in diameter, in the posterior wall of the uterus, which repelled the endometrium and the bladder. The liver, spleen and kidneys were normal. CT and MRI scans were not performed. A day before surgery the neurological examination showed a decrease in the muscular power of the extensor muscles of both feet. Myotonic phenomenon was not found. The patient was thought to suffer from the disease of myotonic dystrophy and not just to carry the gene. Therefore the possibilities for pulmonary complications, malignant hyperthermia, and cardiac arrhythmias had to be kept in mind during the anaesthetic management. After consulting with the patient, written consent was obtained to perform a total abdominal hysterectomy with bilateral salpingo-oophorectomy.

A laparotomy was performed. The abdomen was opened by a midline subumbilical abdominal incision. Exploration of the pelvis revealed the left infundibulo-pelvic ligament stretching inferiorly across the lower third of the uterus from the left to the right, and the uterus to have undergone a 60° rotation along the corpus and the cervix uteri transition-line. Total abdominal hysterectomy with bilateral salpingo-oophorectomy was quickly completed.

The patient’s postoperative course was uneventful and she was discharged in good condition. There were no further urinary difficulties; the patient was healthy on follow-up one month after surgery and started hormone replacement therapy.

The histological report confirmed a large benign myoma measuring 13 x 10 x 8 cm. Also, a small myoma with a maximum diameter of 0.5 cm was found. The fallopian tubes and the ovaries were without pathological significance.

Anesthetic management

Preoperative assessment

The day before surgery a full preoperative assessment was performed. The patient’s height was 1.62 m and her weight 55 kg. She complained of walking disturbances due to the myotonic dystrophy. Heart and pulmonary examination did not reveal any extraordinary organic disturbances. As we mentioned above, in neurological examination the muscular tone and reflexes were reduced in the lower extremities, while the neurological examination of the upper extremities was normal. Information regarding previous anaesthetics (airway and circulatory management or allergies) were negative. Examination of the upper airway system showed good mandibular and neck mobilization in all axes and it was 1-2 by Mallampati class [7]. Laboratory tests for full blood count, SGOT, SGPT, K⁺, Na⁺, prothrombine time, and thyroid function were under normal values. The electrocardiogram (ECG) and the chest roentgenogram were without abnormalities. Blood gas values were normal. Preoperatively, administration of ondasetron (4 mg) as an antiemetic agent and midazolame (0.05 mg/kg) as a conscious sedation were given, both intravenously.

Intra- and postoperative period

On arrival to the operating room, routine intraoperative monitoring was applied: A V5 lead ECG for recording heart rate, a pulse oximeter to control oxygen saturation (SpO₂) and 20-gauge cannula (right radial artery) for continuous blood pressure measurements (systolic and diastolic blood pressure), serial blood gases and haemoglobin determinations. Before the induction of anaesthesia her vital signs were: blood pressure 125/70 mmHg, heart rate 75/min and 97% saturation of O₂. After preoxygenation with 50% oxygen for 3 min the tracheal intubation was facilitated with 110 mg of propofol, 50y of fentanyl and 30 mg of atracurium, all administrated intravenously. The patient was intubated with cuffed tracheal tube without difficulty and then she was ventilated to maintain the end tidal volume of CO₂ concentration in the range between 32 mmHg and 40 mmHg. Also, respiration monitoring included mass spectrometry to measure inspired and expired anaesthetic and respiratory gases. The response to intubation was good and afterwards her vital signs were: blood pressure 130/75 mmHg, heart rate 75 beats per minute and 99% saturation of O₂. Anaesthesia was maintained with 65% nitrous oxide (N₂O) in oxygen supplemented with 1% propofol in a dose of 4 ml/kg/h slowly infused. Neuromuscular blockage agent (10 mg atracurium) was maintained with subsequent incremental doses on the train-of-four (T.O.F.) twitch response. Crystalloids were administered and a urinary catheter was used to quantify urine output as a guide to fluid management. Intravenous fluids were balanced to maintain a urine output of 0.5-1 ml/kg/h. Analgesic demands were covered by giving fentanyl and the doses were lower than those usually administered in order to prevent postoperative respiratory insufficiency, which should be avoided in patients with myotonic dystrophy [8]. The total administration of fentanyl during the operation was 350 mg. Her vital signs were stable and did not exhibit any abnormal variations. Heart function was good without any appearance of cardiac arrhythmias or ECG alterations. Before the end of the operation 10 and 20 mg of pethidine were administrated intravenously and intramuscularly, respectively, for postoperative analgesia. Neuromuscular “monitoring” showed absence of neuromuscular block in 75% 140 min after the induction dose. The extubation was achieved easily when all the reflexes of the patient reappeared and the communication with the environment was acceptable. Neomamine or any other kind of neuromuscular reversal agents were not administrated in order to avoid possible prolonged muscle contractions due to myotonic dystrophy.

In the recovery room the status of the patient after 70 min was stable and the patient did not present any troublesome signs. During this time she was ventilated with 40% oxygen with a Venturi mask and her vital signs were monitored. She did not present any respiratory disturbances and she did not complain of any muscular weakness or pain. The postoperative care after two hours and during the next day was without any pathological alterations.
Discussion

The anaesthetic management of patients with myotonic dystrophy must take into account the possible damage of the disease in multiple organs. A thorough history and physical examination are essential, with special attention to the degree of the present myotonic dystrophy, the amount of muscle wasting, the joint involvement, the pathology within the mouth and the cervical modility, which sometimes is limited by the muscular retraction [3, 4]. Preoperatively, heart function must be evaluated because defects of cardiac conduction and rhythm frequently are present when the heart is affected [3, 4]. Also, pulmonary and thyroid function tests should be performed, together with measurement of arterial blood gases, and finally, a blood ionogram to search for hypokalemia and track down possible diabetes [3, 4].

The choice of the anaesthetic technique and drugs are important for the correct anaesthetic management of a patient with myotonic dystrophy. In our case we used general anaesthesia for total abdominal hysterectomy and preferred the administration of non-depolarizing muscle relaxant (atracurium) in order to prevent muscle contractions and the possibility of difficult ventilation as an adverse effect of administration of succinylcholine [9], as well as the possibility of appearance of malignant hyperthermia [10]. In addition, succinylcholine is not recommended for patients with myotonic dystrophy as this drug can elevate serum potassium, which can aggravate myotonia [4]. In general, non-depolarizing blockers of short or intermediate duration such as atracurium and mivacurium are preferable to pancuronium, which is a longer acting agent [11]. We used 2 mg/kg of propofol which was adequate to induce general anaesthesia deep enough to permit tracheal intubation in our patient. Anaesthesia was maintained at a depth adequate to facilitate controlled ventilation and surgery, by 65% nitrous oxide in oxygen supplemented with 1% of propofol in a dose of 4 ml/kg/h slowly infused. The maintenance in anaesthesia was not done with any kind of volatile agents because of their adverse effects on the myocardium [12, 13].

Finally, the anaesthetic management of a patient with myotonic dystrophy includes follow-up of the immediate postoperative period [3], as muscular atrophy may affect the sternocleidomastoid muscles or the accessory muscles of respiration [4]. A patient with sternocleidomastoid atrophy may not be able to raise her head, and this may lead to respiratory obstruction in the postoperative period. Ventilation may also be impaired because of muscular weakness [4]. Therefore, when the patient awakens, it is necessary to watch for full respiratory autonomy before extubation [3]. In addition, in patients with myotonic dystrophy trouble swallowing during the postoperative period can exist due to pharyngolaryngeal paresis [3]. In our patient the immediate postoperative course of the patient was without any pathological alterations.

Torsion of the non-gravid uterus in women is extremely rare and is more common in animals than in humans [5, 6]. The first axial rotation of the uterus appears to have been reported by Times in 1861 and since then more cases have been reported [6]. Leiomyomas of the uterus are found in a great number of cases [14]. However, torsion of a nongravid, nonmyomatous uterus has been reported. Harvey and Hudson, reported torsion in a 3-year-old girl due to a large hydrosalpinx [15]. In addition, Collinet et al., described a case of torsion of a nongravid uterus due to a large ovarian cyst [5]. In our patient the uterine pathology (large leiomyoma) in combination with the disease of myotonic dystrophy, seemed to be the predisposing factors for the torsion of the non-gravid uterus.

Uterine torsion produces symptoms of varying severity, depending on the degree of the rotation and the speed at which the torsion develops [5]. Abdominal pain is the major symptom [14]. It may vary from mild to acute abdominal discomfort with shock [5]. Other symptoms have also been described such as vaginal bleeding, urinary tract symptoms and gastro-intestinal symptoms [5]. In this case, the only presenting symptom of our patient was her acute weakness to pass urine. Nesbitt and Corner, in a review of 108 cases of torsion of gravid uterus, showed in only one case the feature of interstitial obstruction, although 18.7% had some symptoms related to the gastrointestinal tract, such as nausea, vomiting or diarrhoea [16]. The majority of cases are not correctly diagnosed preoperatively [17]. Diagnosis may take place by a changed position of fibroids seen on previous ultrasound scans or plain radiographs [17]. Both symptomatic and asymptomatic patients require a laparotomy leading to a total hysterectomy or myomectomy [6]. Manual untwisting of the uterus in cases of prolonged torsion with subsequent necrosis and blood thrombosis must be avoided in order to prevent pulmonary embolism [5]. For uncomplicated and carefully selected cases Collinet et al., suggested a conservative surgical approach [5].

In conclusion, we have reported an extremely rare case of a patient suffering from myotonic dystrophy who presented with acute weakness to pass urine due to torsion of a leiomyomatous nongravid uterus. In addition, we presented the anaesthetic management for total abdominal hysterectomy in myotonic dystrophy and discussed the anaesthetic implications in such cases.

References


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