Case Report

Lower leg compartment syndrome following laparoscopic uterine malignancy surgery for uterine cancer complicated by rheumatoid arthritis: a case report and literature review

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Background: Compartment syndrome is a condition in which an increased internal pressure in the upper/lower leg compartment results in circulatory disturbance causing muscle and nerve dysfunction. There have been five case reports of compartment syndrome following gynecologic laparoscopic surgery. We present this rare case herein with a review of the related literature. Case: The patient was a 44-year-old woman. She underwent laparoscopic modified radical hysterectomy for uterine cancer. During surgery, she wore elastic stockings from her toes to her knees. She also had intermittent pneumatic compression of the thighs and lower legs with the head down at 15 degrees. The operative time was 6 h and 49 min. She was diagnosed with compartment syndrome, and a decompression incision was indicated. Conclusion: After experiencing this case, we use the open leg position, releasing the high pelvic position for 10 min every 3 h, during which the nurse should decompress the foot.

Keywords
Compartment syndrome; Laparoscopic surgery; Endometrial cancer

1. Introduction
Compartment syndrome is a condition in which an increased internal pressure in the upper/lower leg compartment results in a circulatory disturbance causing muscle and nerve dysfunction. Although most commonly caused by trauma, compartment syndrome can also be caused by surgical positioning as first described by Volkman et al. [1] in 1882. Later, in 1979, Leff et al. [2] reported a case caused by the lithotomy position. Well leg compartment syndrome (WLCS) is a surgical complication of improper intraoperative positioning [3] that has an incidence of 1/3500, as reported by Halliwill et al. [4]. In addition, a German survey reported a WLCS incidence rate of 0.067% to 0.28% after gynecological surgery [5]. Delayed diagnosis and treatment of WLCS can eventually lead to renal failure, rhabdomyolysis, irreversible nerve damage, and sepsis, which can be fatal [6, 7].

In recent years, endoscopic surgery has been introduced in the gynecology field for treating malignant tumors and prolonged surgeries in the lithotomy position have become more common. There are many reports of WLCS cases after colorectal cancer surgery, which are also performed for the pelvis in the lithotomy position [8]. However, to the best of our knowledge, there have been only five case reports of WLCS following gynecologic laparoscopic surgery including our report [9–12]. The present case is of a laparoscopic modified radical hysterectomy performed for uterine cancer. The surgery was prolonged in the lithotomy position causing postoperative compartment syndrome for which fasciotomy was performed.

2. Methods
We conducted a literature search using a MEDLINE search and Web of science (August 2020). The keywords were compartment syndrome, gynecology surgery, and laparoscopic surgery searched from 1970 to 2020. We selected cases of compartment syndrome after gynecologic laparoscopic surgery. We will also report a case report of our experience.

3. Results
3.1 Case report
The patient was a 44-year-old primigravida woman. At the age of 37, when she visited his local doctor complaining of pain in her left foot, she was diagnosed with chronic rheumatoid arthritis based on bone atrophy in her left foot and blood sampling results. She had a history of taking Methotrexate and Salazosulfapyridine and her condition was stable. She was 163 cm tall, weighed 85 kg, had a body mass index (BMI) of 32.0 kg/m², and had no history of smoking. She visited a gynecologic clinic for abnormal genital bleeding lasting approximately 2 weeks 3 months before being referred to our department.

Endometrial histopathology revealed endometrial carcinoma G1, and uterine cancer was diagnosed. She was referred to our department for further examination and treat-
ment of endometrial cancer. Computed tomography (CT) showed no obvious lymph node or distant organ metastasis, and magnetic resonance imaging results showed that the tumor had invaded into half the depth of the myometrium. Therefore, she was diagnosed with stage IA uterine cancer and underwent laparoscopic modified radical hysterectomy, bilateral adnexal resection, and pelvic lymph node dissection. The surgery was performed under general anesthesia with the patient in the lithotomy position. During surgery, she wore elastic stockings from her toes to her knees to prevent deep vein thrombosis. She also had intermittent pneumatic compression of the thighs and lower legs. She was insufflated through the umbilical region by the closed method with the head down at 15 degrees, and a 5-mm trocar was inserted to observe the abdominal cavity. A 12-mm trocar was inserted into the left lower abdomen, and there were 5-mm trocars in the right lower abdomen, lower midline, and on the right and left umbilical sides respectively. With a total of six trocars, the operation began with a pelvic lymph node dissection. Subsequently, a modified radical hysterectomy and bilateral adnexal resection were performed by cutting the anterior layer of the bladder uterine ligament. Anti-adhesion material was applied onto the entire wound, and the surgery was completed. The operative time was 6 h and 49 min, the blood loss was 9 mL, and the weight of the removed uterus was 257 g. During the operation, insufflation was once suspended for 10 min at 3 h after the start of the operation, and the patient was returned to a horizontal position.

After waking from general anesthesia and extubating, she complained of sudden pain in her left leg. Her left gastrocnemius muscle was markedly swollen, and the distinct pain was localized to this muscle as well (Fig. 1). She had motor deficits, but her skin sensation was normal. Prompt contrast-enhanced CT showed no obvious arterial occlusion or venous thrombus, but tension was observed in all four compartments of the left lower leg (Fig. 2). Following consultation with an orthopedic surgeon, she was diagnosed with compartment syndrome, and a decompression incision was indicated.

A skin incision was made on the medial and lateral sides of the left lower leg extension, the fascia was incised, and decompression of the internal muscles was performed (Fig. 3). Changes in serum creatinine kinase (CK) concentrations are shown in Fig. 4. Immediately after surgery, the CK level was 1089 U/L, which increased to 7861 U/L on postoperative day 1, decreased to 3058 U/L on postoperative day 3, and continued to decline thereafter. In terms of lower extremity motor deficits, she began rehabilitation on postoperative day 2. Before the start of rehabilitation, the manual muscle test for her left lower extremity showed a level of 3–4, and she had numbness in the medial and plantar areas. On postoperative day 3, she could walk with a walker, a cane on postoperative day 4, and with no aide postoperative day 9 onward. Pathological examination revealed that the histological type of the uterine body was endometrial carcinoma G2 with one pelvic lymph node metastasis (1/43) and metastasis to the right ovary. As a result, uterine cancer IIIC1 was diagnosed with a high risk of postoperative recurrence. Carboplatin and paclitaxel were administered as adjuvant chemotherapy. CT after three cycles showed multiple nodules and invasive shadows in the lung fields. Concomitant polymyositis and associated interstitial pneumonia were diagnosed, and chemotherapy was discontinued. She underwent steroid pulse therapy for polymyositis, which required 3 months of inpatient treatment. Her interstitial pneumonia has since abated and she is taking oral steroids on an outpatient basis. In the meantime, there was no deterioration of the left lower extremity after compartment syndrome and chronic rheumatoid arthritis. She is now in postoperative month 34 with no recurrent findings and good postoperative course of compartment syndrome with no evidence of clinical sequelae. For polymyositis, she has been prescribed steroids in the internal medicine department and is currently in stable condition.

3.2 Literature review

A MEDLINE search led to 15 articles. On the other hand, Web of Science led to 5 articles. We reviewed the 20 articles and found 5 reports of compartment syndrome with fasciotomy after laparoscopic gynecologic surgery (including our case). Table 1 shows these 5 cases [8–11].

In a 2009 report, Tomassetti et al. [9] performed laparoscopic surgery in the lithotomy position for endometriosis of r-AFS stage IV in a 30-year-old woman. Her operative time was 480 minutes, and she was postoperatively diagnosed with right lower limb compartment syndrome and underwent fasciotomy. In 2011, Lawrenz et al. [10] reported a 30-year-old woman with a BMI of 25.4 who underwent laparoscopic radical hysterectomy in the lithotomy position for stage IB1 cervical cancer. The operative time was 255 minutes, and the blood loss was less than 100 mL. Postoperatively, she was diagnosed with left lower limb compartment syndrome and performed fasciotomy. In 2013, Boesgaard-Kjer et al. [11] reported a 45-year-old woman with a BMI of 47 who underwent laparoscopic myomectomy in the lithotomy position for fibroid myoma. The operative time was 300 minutes, and
Fig. 2. Contrast-enhanced computed tomography images of the lower extremity. Tension was observed in all four compartments of the left lower leg. Compartment syndrome was diagnosed.

Table 1. Cases of compartment syndrome after gynecologic laparoscopic surgery.

| Year/Reference | Procedure                          | Age (year) | BMI (kg/m²) | Operative time (min) | Blood loss (mL) | Lithotomy position | Intermittent air pressure method | Release from head down position during operation |
|----------------|------------------------------------|------------|-------------|----------------------|----------------|--------------------|-------------------------------|---------------------------------------------|
| 2009 [9]       | Laparoscopic resection of endometriosis | 30         | N/A         | 480                  | N/A            | Y                  | Y                            | N/A                                        |
| 2011 [10]      | Laparoscopic radical hysterectomy   | 30         | 25.4        | 255                  | >100           | Y                  | Y                            | N/A                                        |
| 2013 [11]      | Laparoscopic myomectomy             | 45         | 47          | 300                  | 500            | Y                  | N/A                          | N/A                                        |
| 2016 [12]      | Laparoscopic salpingotomy           | 23         | Morbid obesity | 90                 | N/A            | Y                  | Y                            | N/A                                        |
| 2020 This case | Laparoscopic modified radical hysterectomy | 44         | 32          | 409                  | 9              | Y                  | Y                            | Every 3 hours                              |

BMI, body mass index; N/A, Not available; Y, yes.

Fig. 3. Emergency fasciotomy. A skin incision was made on the medial and lateral sides of the left lower leg extensor, fascia was incised, and decompression of the internal muscles was performed.

the blood loss was 500 mL. Postoperatively, she was diagnosed with bilateral lower limb compartment syndrome and underwent fasciotomy. In 2016 Nicole et al. [12] reported a 23-year-old, morbid obesity woman who underwent laparoscopic salpingectomy for the rupture of a right tubal pregnancy. The operative time was 90 minutes. Postoperatively, she was diagnosed with bilateral lower limbs compartment syndrome and had a fasciotomy.

5. Discussion

Compartment syndrome caused by surgery can be caused by obesity (overweight), hypotension, hypothermia, decreased circulating blood volume, prolonged lithotomy position, pelvic elevation, and external pressure [11]. In pelvic surgery, pelvic elevation reduces blood circulation to the lower extremities, and repositioning may exert pressure on the lower legs [13]. Furthermore, in laparoscopic surgery, pneumoperitoneum causes a decrease in venous return, which reduces blood flow to the lower extremities [8]. These factors are likely to increase the risk of compartment syndrome. Eleven cases of compartment syndrome were reported after laparoscopic colorectal cancer surgery in the lithotomy position that lasted for more than five hours. Nine of them underwent a decompression incision, and two of those cases were followed up [8]. Gynecologic laparoscopic surgery is rarely considered to cause compartment syndrome because it was mostly performed for benign tumors with a shorter operative time. In recent years, however, laparoscopic surgery has been actively performed for
malignancy, and the number of surgeries seems to be increasing. Five reports of compartment syndrome after gynecologic laparoscopic surgery have been documented including this case (Table 1) [9–12]. However, only three cases of compartment syndrome have been reported after open surgery [14–16]. Among surgeries of comparable difficulty levels, open surgery is performed in a shorter time than laparoscopic surgery. Additionally, open surgery does not require high pelvic elevation even in the lithotomy position; therefore, the incidence of this syndrome is considered to be low.

In all five reported cases, laparoscopic surgery was performed in the lithotomy position. The patients were in a relatively higher weight range including the patient who had a BMI of 32 kg/m². The operative time was 6 h and 49 min in the present case. This long operative time was due to the obesity of the patient with a BMI of 32 with a significant amount of fat that caused prolonged lymphadenectomy. In previous reports of the cases in Japan, the operative time was longer than 4 h except for one patient with ectopic pregnancy who developed compartment syndrome at 90 min after the start of the operation [12]. However, the number of reported cases is too small to predict the development of compartment syndrome due to the lack of clear evidence, but prolonged elevation of the pelvis in the lithotomy position is a risk factor. The patient had a history of rheumatoid arthritis, and her vessels may have been more fragile than usual. Rheumatoid arthritis tends to cause vasculitis which is also known as rheumatoid vasculitis, and reported to be most common in the lower limbs [17]. In this case, the patient did not have the systemic condition of rheumatoid vasculitis, but the inflammation and vulnerability of blood vessels may be a factor. The lithotomy position during surgery, intermittent pneumatic compressions, and prolonged surgery with elastic stockings in our patient may have compressed the originally fragile blood vessels in the leg causing them to swell and resulting in the development of compartment syndrome.

The differential diagnosis of compartment syndrome is deep vein thrombosis (DVT), which requires absence of DVT on contrast-enhanced CT after the onset of symptoms as in this case. Risk factors for DVT include prolonged surgery, malignancy, old age, and large pelvic masses [18, 19]. We assessed the present patient to be at a high risk of DVT; therefore, we used elastic stockings and an intermittent pneumatic compression device during the operation; these were our usual measures of DVT prevention. The patient was also given low-molecular-weight heparin for a week after returning to the postoperative room. However, these external foot compressions have also been reported to be involved in the development and aggravation of compartment syndrome [20, 21]. Some reports suggest that it is safer not to use elastic stockings or other intermittent pneumatic compression techniques in lithotripsy surgery [22]. The countermeasures and prevention of DVT and compartment syndrome are contradictory thus making it a challenge to decide which of the two should be the emphasis.

From the above, there is no clear evidence or guidelines for compartment syndrome in laparoscopic surgery for malignancies. However, reports from various authors have said that the greatest risk of compartment syndrome may be the lithotomy position [4, 9, 11]. In our department, after this

**Fig. 4. Changes in serum creatinine kinase concentrations.**
case, the lithotomy position was stopped, and laparoscopic surgery was performed in the open leg position. There is also no exact time frame, but some reports suggest that it is better to release the pelvic elevation in about 2 to 4 hours \[9, 23\]. In our department, the high pelvic position is released for 10 min every 3 h during which the nurse decompresses the feet. However, there is no clear evidence that repositioning is useful to avoid compartment syndrome, and further studies are required to investigate more cases. More than two years have passed since these measures were implemented and not a single case of compartment syndrome has occurred.

6. Conclusions

Complications of vasculitis such as rheumatism, lengthy surgery, and lithotomy position may need to be considered as a risk of compartment syndrome.

Author contributions

HS, YK, and MK were major contributors in writing the article. HS, YK, ST, MO, RS, and KY made clinical examinations, performed surgery, and administered chemotherapy. NM supervised the project. All authors have read and approved the manuscript.

Ethics approval and consent to participate

All subjects gave their informed consent for inclusion before they participated in the study.

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Conflict of interest

The authors declare no conflict of interest.

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