

Recurrent peritoneal inclusion cysts successfully treated with oral contraceptives: a report of two cases

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Summary

Objective: To examine whether conservative treatment with oral contraceptives is effective in the shrinkage of a peritoneal inclusion cyst (PIC). This is a case report of two patients with a PIC that developed after gynecological surgery. **Cases:** Both cases were suspected of a PIC based on the medical history, laboratory data, and image findings. It was difficult to differentiate a PIC from an ovarian tumor. Surgery was chosen at first. However, PICs in both cases recurred after surgery and were treated with oral contraceptives as a conservative treatment. PICs shrank after the treatment of oral contraceptives in both cases. **Conclusion:** Due to the high rate of recurrence following surgery, conservative treatment is recommended to treat PICs. Hormone therapy using oral contraceptives seems to have some therapeutic benefit for the PICs.

Key words: Peritoneal inclusion cyst; Oral contraceptives; Conservative treatment; Sclerotherapy; Vibramycin.

Introduction

A peritoneal inclusion cyst (PIC) is a pseudocyst caused by the diminished ability of the peritoneum to absorb ascites and fluids from the ovaries due to the adhesions around the ovaries caused by pelvic surgery [1-3]. Conservative treatment is often chosen for the treatment of a PIC following surgery, including gonadotropin-releasing hormone antagonist (GnRHa), and hormone therapy using oral contraceptives [1,3-8]. The authors report two cases of recurrence of PIC after surgery, which were effectively treated with oral contraceptives.

Case Report

Case 1

A 46-year-old woman consulted a physician with a complaint of severe lower abdominal and back pains. She had undergone a total abdominal hysterectomy without removing the ovaries due to uterine leiomyoma two years prior. A computed tomography (CT) of the pelvis displayed a multilocular cyst. Magnetic resonance imaging (MRI) revealed a multilocular cystic mass measuring 95 × 60 mm in a diameter. No sign of malignancy was noted. A preoperative diagnosis was a PIC or an ovarian benign tumor. She underwent an exploratory laparotomy (two years and 11 months after initial surgery). Based on intraoperative findings, the patient was diagnosed as having a PIC, and the adhesions were lysed. Her symptoms disappeared after surgery.

One year and four months after surgery (four years and three months after the initial surgery), she complained of lower quadrant discomfort and lower back pains. An 83.9 × 73.3 mm-sized multilocular cyst was found with transvaginal ultrasound. MRI demonstrated a multicystic mass measuring 63 × 90 × 68 mm that showed hypointensity on T1-weighted images and hyperintensity on T2-weighted images (Figure 1). The tumor markers were within normal ranges (CEA: 2.6 ng/ml, CA-125: 11.1 U/ml, and CA19-9: 9.2 U/ml). Based on the history of the present illness, laboratory data,

and image findings, the patient was diagnosed as having the recurrence of PIC. Due to the severity of lower quadrant discomfort and lower back pains, the content of a PIC was transvaginally aspirated with the drained amount of the fluid being 142 ml. The cytology of the fluid was of no malignancy. One month later, however, a PIC expanded up to 42.5 × 50.1 mm in size. Despite her severe symptoms of lower quadrant discomfort and lower back pains, the patient declined further treatment such as oral contraceptives. Thereafter, she was followed up at an outpatient basis, and her symptoms spontaneously disappeared.

Eight months after the aspiration (four years and 11 months after initial surgery), the symptoms developed again. The sclerotherapy using ethanol or GnRHa therapy was thought to be a candidate for the conservative treatment. The patient desired to avoid the side-effects of these therapies. The authors decided to use oral contraceptives since the patient had premenopausal symptoms such as malaise and fatigue, as confirmed by the endocrinological studies showing follicle-stimulating hormone (FSH) of 8.9 mIU/ml and estradiol of 51 pg/ml. The oral contraceptive therapy with Anjyu was begun. The 28-day treatment cycle pack consisted of four dosing phases; the first six days of levonorgestrel 0.05 mg/ethinyl estradiol 0.03 mg, five days with levonorgestrel 0.075 mg/ethinyl estradiol 0.04 mg, ten days with levonorgestrel 0.125 mg/ethinyl estradiol 0.03 mg, and seven days with placebo. After taking oral contraceptives, the PIC shrank to 12.8 × 8.4 mm in size four months later. The patient's menopausal symptoms were abated immediately after the intake of the oral contraceptives. However, seven months after the beginning of oral contraceptives (five years and six months after initial surgery), the PIC grew to 73.9 × 37.5 mm during the continuous intake of oral contraceptives. The patient discontinued the oral contraceptives due to liver dysfunction. The patient did not desire other treatment, and was followed up at an outpatient basis. Nine months after cessation of oral contraceptives (six years and three months after initial surgery), she visited the present hospital with the worsening of the symptoms. Oral contraceptives were resumed again after confirming her recovery from liver dysfunction (Figure 2).

Case 2

A 38-year-old woman underwent a total abdominal hysterectomy and unilateral salpingo-oophorectomy for a tubo-ovarian ab-

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scess. Two years and ten months later, a transvaginal ultrasound revealed a 128 × 109 mm multilocular cyst. MRI showed an ovarian cystadenoma and fluid retention around the cyst, which was suspected of a PIC. An exploratory laparotomy was performed, which revealed an ovarian cyst and a PIC. The ovarian cyst was enucleated, and adhesions were lysed. Due to the severe adhesions, some of the PIC remained. The sites where adhesions were lysed were fixed with ethanol. Pathological diagnosis was a hemorrhagic corpus luteum cyst. A 74 × 53 mm-sized PIC remained after surgery (Figure 3).

Nine months later, she visited to us for a follow-up of the surgery. She complained of lower quadrant discomfort with hot flashes. Transvaginal ultrasound revealed a multilocular cystic mass measuring 86.7 × 51.8 mm in size. With regards to the tumor markers, CA125 levels were slightly elevated up to 37.5 U/ml, but CEA (1.4 ng/ml) and CA19-9 (8.1 U/ml) levels were within normal ranges. The tumor was suspected to be a benign ovarian tumor. One month later, a transvaginal ultrasound revealed the enlargement of a multilocular cyst up to 100 × 66.3 mm (Figures 3 and 4). However, CA-125 levels returned to a normal level of 9.9 U/ml.

She was diagnosed as having the exacerbation of a remaining PIC. She was treated conservatively. The hot flashes were abated

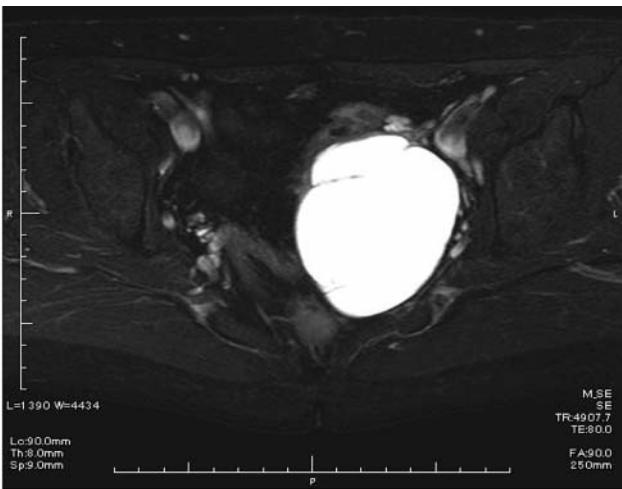


Figure 1. — Case 1, MRI, T2-weighted, fat suppressed images, showed a multicystic mass of hyperintensity measuring 63 × 90 × 68 mm.

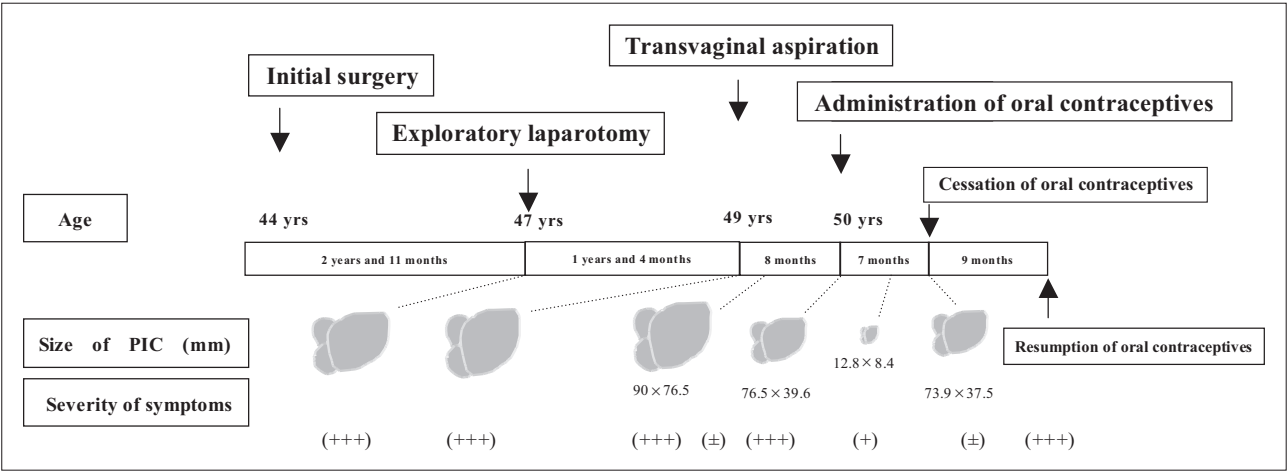


Figure 2. — Chart of the course of case 1.

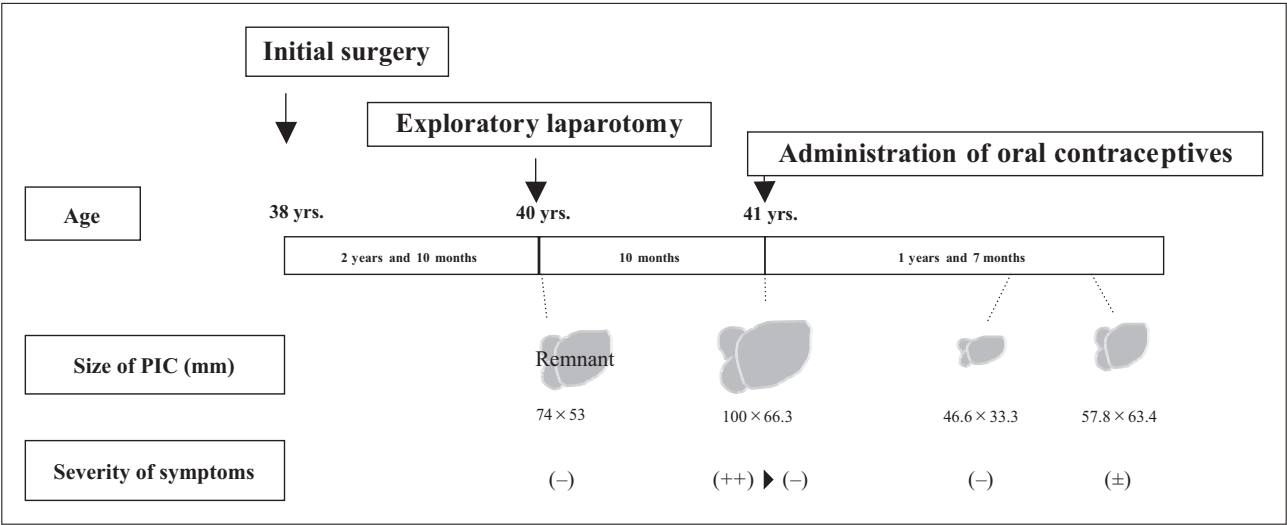


Figure 3. — Chart of the course of case 2.

with treatment with Chinese herbal medicines. However, menopausal symptom of chill remained, and the treatment with oral contraceptives (Anjyu) was begun. Serum levels of FSH were 9.2 mIU/ml and estradiol of 55 pg/ml. Immediately after the intake of contraceptives, lower quadrant discomfort and menopausal symptoms with chills disappeared and the remaining PIC shrank. One year and one month after the initiation of oral contraceptives (four years after the initial surgery), the PIC shrank to 46.6×33.3 mm (Figure 3). The patient has continued to take oral contraceptives, and the size of the PIC is of 57.8×63.4 mm (Figure 3).

Discussion

A PIC is a pseudocyst caused by the diminished ability of the peritoneum to absorb ascites and fluids from the ovaries due to the adhesions around the ovaries caused by pelvic surgery [1-3]. A PIC develops with an inflammation of the peritoneum as a result of pelvic surgery, infection, and endometriosis [1,2,4]. Capillaries and lymph ducts located throughout the peritoneum first become occluded and diminish the ability of peritoneum to absorb fluid [9,10]. Then, ascites are enclosed by the peritoneum, and the fluid is retained in those cavities instead of being absorbed [9,10]. The PIC often develops on the left-side of the pelvic cavity, since the broad mesentery of the sigmoid colon is extensively involved in the formation of closed cavities [4]. Retained ascites is mostly considered to be inflammatory exudate or fluid from the ovaries [1]. Especially when the ovary is enclosed with a closed cavity, secretions from the ovarian surface, blood from ovulation, or follicular fluid also flow into the closed cavity and helps the PIC to grow [6].

A PIC is one type of cystic mass within the pelvic cavity that needs to be differentiated from other masses. Establishing a diagnosis based on clinical manifestation and image findings is crucial to determine the treatment strategy. This is because PICs have a high rate (30-50%) of recurrence following surgery [2,4,9]. The ovary coming into contact with a closed cavity is a mechanism for the onset of a PIC [11]. Evidence of this contact is important for diagnosing a PIC [11]. Clinical manifestations and image findings of the PIC include: (1) a previous history of gynecologic surgery, endometriosis, and pelvic peritonitis, (2) the presence of an active ovary, (3) symptoms such as lower abdominal pains and lower abdominal discomfort, (4) image findings of a cystic mass with irregular margins and no solid component which fills the space inside the pelvic cavity, the ovaries located inside or outside of the margins of the mass, and (5) tumor markers within the normal range [11,12]. Once the diagnosis of the PIC is established, conservative treatment is recommended, given the high rate of the recurrence following surgery [9]. Surgical treatment should be adapted only to patients in whom PICs are hard to differentiate from ovarian tumors, those with large PIC lesions, and those with severe symptoms.

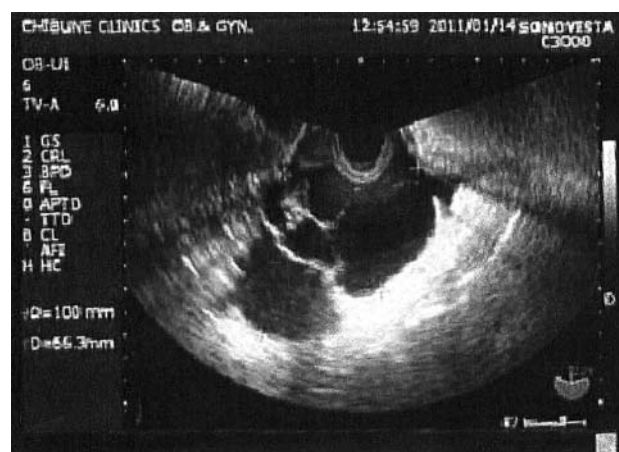


Figure 4. — Case 2: transvaginal ultrasound image revealing enlargement of a multilocular cyst up to 100×66.3 mm.

In the present cases, two patients had severe symptoms, and the differentiation between a PIC and an ovarian tumor was difficult. Surgery was chosen initially to treat PICs in both cases. In the first case the PIC disappeared with the lysis of adhesions but recurred afterwards, while in the second case a part of the PIC remained. The present cases also confirmed the high rate of recurrence following surgery and the difficulty in surgical procedure itself.

Aspiration is another choice to treat the PICs [13]. Sclerotherapy using vibramycin (doxycycline) or ethanol was shown to be effective when performed with aspiration [7,14,15]. The disappearance or shrinkage of PICs and a lower rate of recurrence in comparison with surgery have been reported with the combined therapy of sclerotherapy using vibramycin (doxycycline) or ethanol, whereas aspiration alone shows a similar recurrence rate to that of surgery [5,16]. In the first case, only aspiration was done to treat recurrence of the PIC, but a repeated recurrence occurred. In the second case, sclerotherapy with ethanol was combined with surgery, and recurrence did not occur in the lesion where sclerotherapy was performed. However, the remaining PIC became a problem later. When surgery is performed, combining sclerotherapy with surgery or aspiration could presumably curb the recurrence rate and increase the patient's potential to recover.

Conservative treatment includes GnRHa therapy and hormone therapy using oral contraceptives. Currently, the response rate to hormone therapy using oral contraceptives is still remains unclear [1,6-8,17]. Oral contraceptives suppress ovulation, preventing fluid retention by a decreased fluid production [1,5]. In both cases, the size of the PIC fluctuated during the treatment with oral contraceptives. Therefore, oral contraceptives cannot be categorically described as having sufficient effects on the shrinkage of PICs. Nonetheless, it was noticed that the PIC did not grow remarkably after the beginning of oral contraceptives. In the first case, the symptoms were not completely alleviated de-

spite the shrinkage of the PIC, but surgical treatment was not required (Figure 2). In this case, after cessation of oral contraceptives, severe subjective symptoms recurred. Thus, oral contraceptives have some therapeutic benefit as a conservative treatment for PICs in addition to an auxiliary benefit of treating menopausal symptoms.

An advantage of GnRHa therapy is its potential therapeutic benefit. Several reports have indicated that GnRHa therapy eliminates PICs and involves little recurrence rates [7,17]. GnRHa therapy should be the first choice when conservative treatment is chosen in patients with the PIC. The add-back therapy using contraceptives with GnRHa agonists is recommended to the case of patients with PICs and menopausal symptoms. After confirming the disappearance of PICs by GnRHa therapy, oral contraceptives may be used as a maintenance therapy. This treatment strategy may further prevent the recurrence of PICs.

Collectively, sclerotherapy using vibramycin (doxycycline) or ethanol is recommended for patients with a PIC as a surgical option, whereas conservative treatment with GnRHa therapy is a first choice. The present cases treated with oral contraceptives for the PICs suggest that oral contraceptives may also be another choice as the conservative treatment for the PICs.

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