CASE REPORT

Polycystic ovary syndrome with asynchronous bilateral adnexal torsion in a natural cycle

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SUMMARY

Cases involving polycystic ovaries (PCOs) with adnexal torsion in a natural cycle without ovulation induction are rare, and no reports of such cases have described asynchronous bilateral adnexal torsion. Here, we report a case of PCO syndrome (PCOS) with asynchronous bilateral adnexal torsion in a natural cycle. The patient was a 37-year-old woman with a history of 2 gravidas 1 para. Her primary complaint was left lower abdominal pain. Ultrasonography and MRI identified multiple uterine myomas occupying the pelvis and the left ovary, with oedematous swelling that had moved ventrally to the uterus. She was diagnosed with adnexal torsion and underwent emergency laparoscopic adnexectomy. Nine months after surgery, she experienced right lower abdominal pain. Ultrasonography revealed suspected right adnexal torsion and she underwent emergency surgery. The right ovary was twisted 540° counterclockwise and swollen to 7 cm in size, with partial polycystic changes. She was histopathologically diagnosed with a PCO, and the final diagnosis, which also considered the endocrine test results, was PCOS. In PCOS, adnexal torsion may occur if the swollen ovary moves because of a hystero myoma or other cause. Accordingly, torsion should be considered during the follow-up of patients with PCOS.

BACKGROUND

Adnexal torsion is mainly thought to be caused by an ovarian tumour and is considered an important acute abdominal gynaecological disease. Polycystic ovary syndrome (PCOS) causes ovulation disorders and the syndrome is frequently encountered in fertility practices. Torsion of the appendage pedicle occurs in approximately 7% of cases of ovarian

hyperstimulation syndrome induced via ovulation induction in normal ovaries.¹ Among PCOS cases, however, reports of the development of torsion of the appendage pedicle during a natural cycle have been rare. Here, we report a case of asynchronous bilateral adnexal torsion in a patient with PCOS without ovulation induction.

CASE PRESENTATION

A 37-year-old woman with a history of 2 gravida and 1 para visited the Department of Internal Medicine at our hospital with a primary complaint of left lower abdominal pain. Her menstrual history was a 60–90 day cycle. She had no significant medical history and there was no family history. Plain CT identified multiple uterine myomas and ovarian swelling, leading to a suspicion of gynaecological disease and referral to our department. Vital signs were within normal range (body temperature, 37.1°C; blood pressure, 142/109 mm Hg and pulse rate, 76/min). The abdomen was flat and soft, with tenderness accompanied with muscular defence in the left lower abdomen. During a pelvic examination, the uterus was found to be anterverso flexion with a size similar to that of a child's head, and a palpable elastic mass accompanied by tenderness was found slightly to the left of the uterus. Transvaginal ultrasonography found multiple uterine myomas occupying the pelvis but could not identify either of the ovaries. No ascites was found in the Douglas pouch. Transvaginal ultrasonography found a 6×2 cm mass that was consistent with the point of tenderness in the left lower abdomen. There was no definite cystic change (figure 1). No enhanced inflammatory reaction was observed (white cell count, 8700/μL; C reactive protein, 0.05 mg/dL), and no abnormal peripheral haematology or blood chemistry results were obtained. Plain pelvic MRI showed that multiple uterine myomas were found to occupy the pelvis. The left ovary, which was 6×2 cm in size, exhibited oedematous swelling located superior-anterior to the uterus with partial cystic changes (figure 2). The right ovary was 2×3 cm in size, without oedematous changes. Although her vital signs were normal, the pain increased and caused vomiting. She underwent emergency laparoscopic surgery for a diagnosis of torsion of the left appendage pedicle.

At abdominal exploration, pelvis was occupied by multiple uterine myomas. The left ovary was swollen to approximately 7 cm in size anterior to the uterus and twisted 540° counterclockwise (figure 3). Although the right ovary was not twisted

Figure 1 Transabdominal ultrasonography. A 6×2 cm mass was observed at the site of tenderness.

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Figure 2 Plain pelvic T2-weighted MRI images: (A) sagittal section and (B) horizontal section. Multiple uterine myomas were identified, and the left oedematous ovary (arrow) had shifted superior-anterior to the uterus. The right ovary was normal in size.

or swollen, it contained multiple small cysts that were presumed to be ovarian follicles. The swollen ovary was oedematous and dark black-purple in colour. Because the colour did not improve even after removing the torsion, a left adnexectomy was performed. Although no evident neoplastic lesion was observed, some cysts were as large as several millimetres (*figure 4*). The surgical duration was 1.5 hours, and 15 g of blood were lost; the left appendage weight was 34 g. The histopathological properties of PCOS, which include thickening and proliferation of the theca interna cell layer and interstitial cell hyperplasia, were difficult to observe in this case because of the bleeding. However, the ovarian parenchyma contained a number of ovarian follicles measuring 2–7 mm in size, leading to a diagnosis of polycystic ovary (*figure 5*). There were no necrotic changes. The patient's postoperative progress was good, and she was discharged on day 3 postoperatively. The results of endocrine tests conducted in an outpatient setting indicated a luteinising hormone (LH) level of 8.1 IU/mL and follicle-stimulating hormone (FSH) level of 8.1 IU/mL. The patient exhibited both oligomenorrhoea and a polycystic ovary and thus met the diagnostic criteria of PCOS².

Figure 3 Image of the abdominal cavity. The left ovary was enlarged to approximately 7 cm in size and had moved ventrally to the uterus. It was twisted 540° counterclockwise.

Figure 4 Image of the abdominal cavity. No definite tumour was observed in the left ovary, although small cysts measuring several millimetres in size were detected.

Eight months after the operation, she developed right lower abdominal pain and visited the Department of Gynaecology in our hospital. Abdominal palpation identified an area of marked tenderness with rebound tenderness in the right lower abdomen. CT observed multiple myomas occupying the pelvis and a 78×65×34 mm mass with uneven internal absorption anterior to the uterus (*figure 6*). The diagnosis was torsion of the right appendage pedicle; 20 hours later, the patient underwent laparotomy. The right appendage was twisted 720° and was dark black in colour due to impeded blood circulation. The oedematous ovary was located anterior to the uterus. Even after removing the torsion, the blood flow did not improve and an adnexectomy was performed. The right appendage weighed 80 g. The ovary was dark brown with marked congestion and partial rupture. Histopathology revealed multiple ovarian follicles, leading to a diagnosis of a polycystic ovary. The preoperative Menstruation was irregular, and the hormone test results indicated LH dominance, with a LH level 5.2 mIU/mL, FSH 2.8 mIU/mL, estradiol 226 pg/mL and testosterone 0.28 mg/mL; the case therefore appeared to indicate PCOS. Her postoperative progress has been good, and she is currently undergoing hormone replacement therapy in an outpatient setting.

DISCUSSION

PCOS occurs relatively frequently in clinical settings and presents with distinctive cardinal clinical symptoms (menstrual disorder due to ovulation disorder, infertility, hypertrichosis and obesity), characteristic morphological changes in the ovaries (ie, polycystic changes) and abnormal incretion (blood LH ≥ FSH, hyperandrogenism). This condition develops in 6.5%–8% of women, according to the often-used Rotterdam criteria.² The patient in the present case presented with oligomenorrhoea and multiple ovarian follicles ranging in size from 2 to 9 mm according to a histopathological assessment, leading to a diagnosis of PCOS.

Adnexal torsion represents an exemplary gynaecological acute abdomen. Tenderness around the appendages during pelvic examination and ultrasonographic findings of ovarian swelling are important to a diagnosis. Torsion of the pedicle tends to occur with benign tumours rather than malignant tumours; the latter has a reported incidence of approximately 2%.^{3,4} Furthermore, torsion rarely occurs in cases involving endometrial cysts

Figure 5 Histopathological image (H&E staining; magnification). Many ovarian follicles measured 2–7 mm in size.

or malignant tumours because of adhesions to surrounding tissues or invasion. Many articles reported that ovarian tumours without adhesion tend to induce torsion of the pedicle on reaching a size of ≥ 5 cm.⁵ In patients with PCOS, torsion of the appendage pedicle is rare during a natural cycle without ovulation induction,⁶ and no previous reports have described asynchronous bilateral adnexal torsion in such cases. Although the present case did not involve a tumour, the ovaries were swollen to approximately 7 cm in size at the time of surgery, despite possible oedematous changes caused by torsion. Therefore, ovarian swelling due to PCOS may have been one of the causes of adnexal torsion.

Adnexal torsion, which is associated with ovarian movement, is more likely to occur during pregnancy than during non-pregnant periods, and torsion of the ovarian tumour pedicle is likely to occur in the first trimester (particularly weeks 10–17), when the ovaries move from the pelvis to the abdominal cavity. Yen and colleagues reported that 60% of cases of pregnancy-related torsion of the pedicle developed during weeks 10–17 of gestation, whereas only 5.9% developed during week 20 and thereafter.⁷ Ovarian movement was also found to be an important diagnostic indicator of torsion of the appendage pedicle.⁸ In the present case, multiple uterine myomas occupied the pelvis, and the ovary had moved anteriorly to the uterus in both the first incidence of torsion involving the left ovarian pedicle and

the second incidence involving the right ovarian pedicle. Interestingly, although the right ovary was posterior to the uterine appendage pedicle during the first operation for torsion of the left ovarian pedicle, the right ovary had become twisted anteriorly to the uterine appendage pedicle by the time of identification of the second torsion of the pedicle. In the present case, multiple uterine myomas led to enlargement of the uterine appendage pedicle to the size associated with week 16 of gestation; therefore, the ovary was likely able to move easily. The bilateral adnexal torsion observed in the present case could be attributed, at least in part, to mild ovarian enlargement consequently to PCOS and uterine enlargement caused by multiple uterine myomas.

Bilateral torsion of the appendage pedicle is very rare, accounting for only 5% of cases of torsion of the pedicle.⁸ Many such cases have been reported in premenarchal girls. Girls in this population may develop torsion of the pedicle, regardless of ovary size, because they have longer utero-ovarian ligaments and a higher level of appendage mobility.^{9–10} Dannhoo and colleagues reviewed 20 cases of bilateral torsion of the appendage pedicles, including 15 cases with synchronous onset and 5 cases involving asynchronous onset.¹¹ Although an adult case involving synchronous bilateral torsion of the ovarian tumours was reported, no bilateral torsion of the appendage pedicles has been reported in cases of PCO without ovulation induction.¹²

In the present case, she underwent CT as an initial study to exclude appendicitis by internal medicine, and then underwent MRI study for differential diagnosis of solid oedematous mass. MRI and CT may be helpful if findings on ultrasound are equivocal, but the cost and time required for MRI and CT do not justify its routine use for the evaluation of suspected adnexal torsion.^{12–13} Ultrasound is less expensive than CT and MRI, and its diagnostic performance is similar. Pelvic ultrasound and Doppler ultrasound are the initial imaging studies of choice for patients with suspected adnexal torsion. A prospective study of patients with adnexal mass in the setting of pelvic pain, decreased or absent venous flow on Doppler was found to have 100% sensitivity and 97% specificity for a diagnosis of ovarian torsion.¹⁴

In the present case, despite removal of the torsion, the shift in left ovarian colour to a dark black-purple hue suggestive of ischaemia did not improve, and a left adnexectomy was therefore performed. However, pathology revealed no evidence of necrosis. Oelsner and colleagues reported that despite the 'necrotic' appearance of the twisted ischaemic ovary, ovarian function is preserved in 88%–100% of cases.¹⁵ Recently, conservative management by detorsion has been reported.^{8–16} Torsion due to an ovarian cyst could prevent the recurrence of adnexal torsion by cystectomy, however, torsion due to PCOS has a risk of recurrence of adnexal torsion, because bilateral swelling ovaries remain. Bilateral oophoropexy could prevent the risk of adnexal torsion. The technique of oophoropexy consists of suturing the ovary to the uterine serosa, the broad ligament or the pelvic side wall using a non-absorbable material.^{8–17} Nagel *et al*¹⁸ described the laparoscopic plication of the utero-ovarian ligaments for the prevention of the twisting recurrence. From the viewpoint of ovarian function preservation, in the present case, conservative management should have been considered at the second surgery.

Because 80% of cases of adnexal torsion occur in women of reproductive age, a diagnosis and treatment should be provided as soon as possible, and appendage-conserving therapy involving the removal of torsion of the pedicle should be the first treatment option.

Figure 6 Enhance CT image. The right ovary had moved anteriorly to the uterus and was swollen to a size of 7×6×3 cm (arrow).

Learning points

- Polycystic ovary syndrome (PCOS) is frequently encountered in clinical settings. Cases involving ovarian swelling or movement in association with uterine myoma or other causes should be followed up in consideration of possible torsion of the pedicle, even if no tumour is present.
- Despite the 'necrotic' appearance of the twisted ischaemic ovary, ovarian function is preserved; rapid diagnosis and provision of treatment as soon as possible are preferred steps with regard to appendage conservation.
- Torsion due to PCOS had a risk of recurrence of adnexal torsion, because bilateral swelling ovaries remain. Bilateral oophoropexy could prevent the risk of adnexal torsion.

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