



Significant elevation in serum CA 125 and CA 19-9 levels with torsion of the hydrosalpinx in a postmenopausal woman

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Isolated torsion of the fallopian tube in postmenopausal women is rare. In this case report, we detail the case of a 53-year-old patient who presented with adenomyosis and a left hydrosalpinx with high levels of CA 125 and CA 19-9. The isolated torsion of the left hydrosalpinx was observed during the operation. The serum levels of CA 125 and CA 19-9 were reduced from 129.62 and 348 to 58.2 and 12.41 U/mL, respectively, after total laparoscopic hysterectomy with salpingectomy. On radiologic evaluation, there were no other factors that may have influenced the increase in serum levels of CA 125 and CA 19-9 in this patient, which were reduced after operation. To the best of our knowledge, this is the first case of association between perioperative changes in CA 19-9 levels and isolated torsion of the fallopian tube.

Keywords: CA 125 antigen; CA 19-9 antigen; Fallopian tubes; Isolated tubal torsion

Introduction

An isolated torsion of the fallopian tube refers to torsion of the fallopian tube that is not associated with any ovarian abnormality. It is extremely rare and occurs in 1 out of 1.5 million women [1]. It is mostly observed in the reproductive age group with rare exceptions in the pediatric and postmenopausal age groups [2]. Furthermore, there has been no reported case to date that describes isolated fallopian tube torsion with hydrosalpinx and adenomyosis in postmenopausal women.

CA 125 and CA 19-9 are used as tumor markers for various malignancies and are observed to increase in other non-malignant conditions as well. However, only a few reports have included the association of adnexal torsion with CA 125 and CA 19-9. To the best of our knowledge, the comparative evaluation of CA 125 and CA 19-9 levels has not been investigated between the preoperative and postoperative conditions in cases of isolated fallopian tubal torsion.

We report the first case with isolated torsion of the fallopian tube demonstrating significant changes in the serum levels of CA 19-9 after salpingectomy in a postmenopausal woman.

Case report

A 53-year-old postmenopausal woman presented with vaginal bleeding that occurred three times within three weeks and the amount was more than 2 well-soaked pads per day. She also expressed abdominal discomfort in the left lower quadrant and developed leg cramps during this period. She was nulliparous and had attained menopause 10 years ago. She underwent the right salpingectomy due to right tubal pregnancy 15 years ago, and was aware of her adenomyosis that

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occurred six years ago. There was no familial history of cancer, particularly ovarian, peritoneal, or fallopian and breast cancer, and the patient had no history of cancer. Contrast-enhanced abdominopelvic computed tomography (CT) was performed for the evaluation of hematuria 1 year ago, which did not show any significant findings related to hydrosalpinx, except for the presence of a left ureteric stone with mild hydronephrosis. She had no other significant past medical and operative histories.

During the physical examination, no tenderness or rebound tenderness was tested in any abdominal regions. Furthermore, there were no abnormal findings on physical examination of the breast. Results of laboratory evaluations including hematologic, urinary, and routine chemistry tests were within the normal range. The serum levels of CA 125 and CA 19-9

were elevated as 129.6 and 348.0 U/mL, respectively. Sonographic examination showed uterine enlargement with heterogeneity of the entire myometrium and presence of a 5.2×3.9-cm-sized tubular cystic lesion in left adnexa. Using abdomen sonographic examination to determine possible causes of elevation of CA 19-9 levels, several possible medical findings influencing the elevation of serum CA 19-9 level were not visible. A preoperative diagnosis was adenomyosis with left hydrosalpinx. Laparoscopic surgery was performed. Intraoperative findings were as follows: a left dilated fallopian tube was measured as 3×9 cm and its dark-blue distal portion was twisted (Fig. 1). There was pelvic peritoneal adhesion around the left round ligament. The uterus revealed the size of a pregnant uterus at 12 weeks. The bilateral ovaries were normal in appearance and the right adnexa was not visible because of salpingectomy for right tubal pregnancy. The left fallopian tube recovered its color gradually after detorsion of the tube was harvested. A laparoscopic total hysterectomy with left salpingectomy was performed.

The pathology report revealed an 8.5×2.2-cm-sized hydrosalpinx and 11.5×7.0×5.5-cm-sized adenomyosis of the uterus. There were no features observed of a congenital abnormality of the fallopian tube. The final diagnosis was the isolated torsion of left hydrosalpinx with adenomyosis. There were no abnormal findings on postoperative gynecologic sonography. The patient recovered from preoperative symptoms with an uneventful postoperative period and was discharged 5 days later. On the 8th postoperative day, the serum levels of

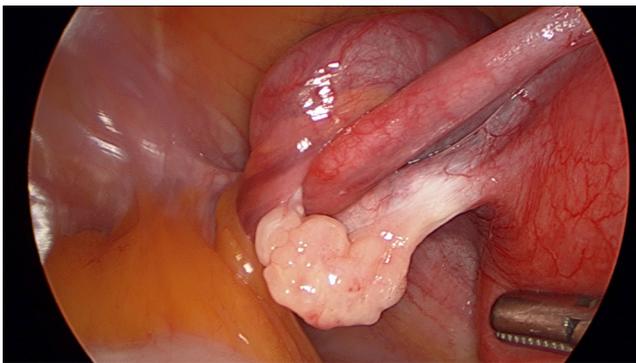


Fig. 1. Laparoscopic view shows the isolated torsion of the left hydrosalpinx, with the normal ovary visible.

Table 1. Clinical characteristics of isolated fallopian tube torsion cases in postmenopausal women

Author	Year	Age (yr)	Age at menopause (yr)	Presenting symptom	Pathologic findings	Tumor markers	Side of torsion	Procedure	Further treatment needed
Ding et al. [5]	2007	70	Unknown	Lower abdominal pain	Hydrosalpinx	Normal ^{a)}	Right	Laparoscopy Salpingo-oophorectomy	None
Toyoshima et al. [2]	2015	63	53	Lower abdominal pain	Paratubal cyst	Unknown	Right	Laparoscopy Paratubal cystectomy Salpingectomy	None
Feng et al. [6]	2012	76	49	Lower abdominal pain	Post BTL status Hydrosalpinx Hematosalpinx	Unknown	Right	Laparoscopy Salpingo-oophorectomy	None
Ozgun et al. [4]	2007	55	50	Lower abdominal pain	Paratubal cyst	Unknown	Right	Total abdominal hysterectomy Bilateral salpingo-oophorectomy	None

BTL, bilateral tubal ligation.

^{a)}The performed tumor marker was not clear, however, the authors mentioned that the tumor markers were normal.

CA 125 and CA 19-9 were reduced to 58.2 and 12.41 U/mL, respectively.

Discussion

Isolated torsion of the fallopian tube without an ovarian abnormality is an uncommon event, occurring mostly during reproductive years and rarely in adolescents [2]. The event is also extremely rare in the postmenopausal women, presumably because of hypotrophy of the fallopian tube and its blood supply [3]. Four cases of isolated fallopian tube torsion in postmenopausal woman were reported to date (indexed in Medline/PubMed). The contents of each case report are summarized in Table 1 [2,4-6]. Furthermore, there has been no report on the association between torsion of the hydrosalpinx with adenomyosis and perioperative changes in serum CA 125 and CA 19-9 levels.

The possible causes of isolated torsion of the fallopian tube are classified into intrinsic and extrinsic factors [7]. Intrinsic factors include congenital tubal abnormalities, acquired pathology (hydrosalpinx, hematosalpinx, tubal neoplasms, and tubal surgery), as well as autonomic dysfunction. Extrinsic factors include ovarian or tubal masses, pelvic adhesions, uterine enlargement, movements or trauma to the pelvic organ, and pelvic congestion. Therefore, in the current case, hydrosalpinx, pelvic peritoneal adhesion, and uterine enlargement related to adenomyosis may have collaboratively contributed to the pathogenesis.

Isolated torsion of the left fallopian tube as described in the current case is rarer than the right side [4]. Many reports suggest that isolated fallopian tube torsion is commonly discovered found on the right side, possibly because of the partial immobilization of the left tube by its proximity to the sigmoid mesentery. Moreover, torsion on the right side is more frequent as the right lower quadrant pain is more often surgically explored secondary to the concerns of appendicitis [3].

The clinical presentation of the isolated fallopian tube torsion is non-specific. The most specific symptom is lower abdominal pain or flank pain, projecting to the side of tubal torsion [2,4]. On physical examination, clinical findings include abdominal tenderness with or without peritoneal signs [2,7]. In the current case, the patient experienced abdominal discomfort but no tenderness in the left lower quadrant.

The sonographic findings of tubal torsion represent a

dilated tube with thickened and echogenic walls [8]. It is helpful when a whirlpool sign that is specific to adnexal torsion is seen [9]. Doppler study may show a high impedance representing the absence or reversal of the diastolic flow in tubular structure [10]. However, in the current case, the typical sonographic findings of the isolated fallopian tube torsion were not visible. The CT findings include an adnexal mass separate from the tube, a twisted appearance of the fallopian tube, tube dilation of greater than 15 mm, a thickened and enhancing tubal wall and luminal CT attenuation greater than 50 HU (Hounsfield units) consistent with hemorrhage [11]. Hence, magnetic resonance imaging may provide valuable information such as tube thickening, ascites and uterine deviation to the twisted side [12]. Unfortunately, CT and magnetic resonance imaging were not performed in this case.

Laparoscopy is the gold standard in the diagnosis and treatment of isolated fallopian tube torsion. In the reproductive age group, laparoscopic tubal detorsion can be considered to preserve fertility for the patient without ischemic damage [3]. However, in cases with presence of gangrenous tissue in the tube, suspicious malignancy, or the woman has completed her family, a laparoscopic salpingectomy is preferred [5].

CA 125 and CA 19-9 are considered to be markers for malignant tumors, and are widely used in clinical practice [13]. CA 125 is studied as a marker of non-mucinous epithelial ovarian tumors; however, CA 125 is not a tumor-specific antigen and is also elevated in patients with endometriosis, adenomyosis, leiomyoma, and pelvic inflammatory disease [14]. CA 19-9 level is frequently increased in gastrointestinal or pancreatic cancer, other malignancies, and in some benign conditions, such as mature cystic teratoma of the ovary [13]. Besides, CA 19-9 has been described as a good marker, particularly for ovarian torsion and the extent of ovarian necrosis [14]. In a retrospective study, which evaluated 163 women who underwent surgery for mature cystic teratoma of the ovary, serum CA 19-9 levels was found to be a useful in the predictive marker of ovarian torsion [15]. In other reports, normalized CA 19-9 levels were observed after surgery due to ovarian torsion [13,14]. In the present case, the preoperative levels of CA 125 and CA 19-9 were elevated and definitely reduced levels after surgery. Elevated CA 125 levels could be potentially influenced by adenomyosis; however, there has been no report on elevated CA 19-9 levels in adenomyosis or hydrosalpinx. In this case, radiologic findings that may influence elevation of serum levels of CA 19-9 were not found

and the level was normalized after hysterectomy with salpingectomy. Hence, it may be suggested that the elevation of serum CA 19-9 on the isolated fallopian tube torsion is due to an inflammatory or ischemic reaction after the torsion.

There has been no report describing the isolated torsion of hydrosalpinx with adenomyosis in the postmenopausal age group. To the best of our knowledge, this is the first report to date describing isolated fallopian tube torsion with elevation of CA 19-9 level, which returned to normal levels after salpingectomy and hysterectomy in a postmenopausal woman. Thus, the association may be suggested between the isolated fallopian tube torsion and elevation of CA 19-9. Further studies will be necessary to assess the perioperative CA 19-9 level when isolated fallopian tube torsion is suspected. It is helpful to investigate the utility of CA 19-9 levels that could be available in the prediction of isolated fallopian tube torsion.

Conflict of interest

No potential conflict of interest relevant to this article was reported.

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