Bilateral Torsion of the Normal Ovary and Oviduct in a Young Girl

May Wakamatsu, MD, Paul Wolf, MD, and Kurt Benirschke, MD
San Diego, California

This report describes the torsion of a normal ovary and oviduct in a $10\frac{1}{2}$-year-old girl. Hemorrhagic infarction of the ovary and fallopian tube occurred, which required surgical removal. This same circumstance had occurred four years earlier on the contralateral side and had resulted in the surgical removal of the opposite adnexa. This condition has been reported previously 16 times. When such torsion occurs the first time, it is important to be aware of the possibility of recurrence and consider the wisdom of foreshortening the opposite mesosalpinx, thus preventing future torsion of the remaining adnexa that would result, as in this case, in sterilization.

When torsion occurs on the right side, as is more common, the clinical diagnosis is usually acute appendicitis. If it occurs on the left side, pelvic inflammatory disease or colonic or ureteral disease is usually suspected, with a delay in accurate diagnosis and treatment.[1] Torsion of ovary and tube is rarely diagnosed in young girls before surgery.

CASE REPORT

A $10\frac{1}{2}$-year-old girl had a history of chronic, intermittent lower abdominal pain and was found to have a pelvic mass. Her history is significant in that she had a left salpingo-oophorectomy in April of 1982 at Children's Hospital, Boston, for torsion of the left adnexa. Since that time she has had chronic abdominal pain, intermittently, which was attributed to the previous surgery. Most recently she presented to the emergency room at the University of California, San Diego, Medical Center because of severe abdominal pain. A sonogram revealed a 9 × 6-cm pelvic mass. The workup included intravenous pyelography, which showed normal findings except for an indentation of the bladder caused by the mass. Magnetic resonance imaging was consistent with the sonographic findings and suggested the presence of either an ovarian dermoid tumor or a distended uterus. The chest radiograph was normal; those of the abdomen did not contribute to the diagnosis. β-Human chorionic gonadotropin level was 3 IU/L (3 mIU/mL) and α-fetoprotein was less than 11.7 μg/L (11.7 ng/mL). Hematocrit was 0.36 (36.2 percent). She had a normal white cell count and differential. The sedimentation rate, electrolytes, and other chemistry determinations were all normal.

An exploratory laparotomy on November 19, 1986, and examination under anesthesia showed a normal vagina and closed cervical os. In the abdomen a 4 × 5-cm necrotic, hemorrhagic mass was found, which represented the right fallopian tube and ovary. The oviduct had undergone torsion at midlevel; there was no ovarian tumor. Uterus was appropriate for her age, and no other abdominal abnormalities were seen. The left adnexa was found absent. Salpingo-oophorectomy was done. She had a normal postoperative course except for mild temperature elevation as a result of urinary tract infection, which was successfully treated with penicillin.

The 1982 specimen was a bluish-black fallopian tube 5 × 1 cm to which was attached a cystic ovary measuring 4 × 5 cm with a smooth surface. When sectioned, it was found to contain a blood clot. Microscopically the tissues were infarcted and hemorrhagic. Primary follicles could be identified; there was no tumor, however. The specimen of the 1986 operation showed similar features. Tube and ovary had undergone hemorrhagic infarction, and histologically only a very thin layer of ovarian cortex could be recognized with some primary follicles; again there was no tumor. It is, of course, possible that a hemorrhagic ovarian cyst was the cause of the torsion, rather than its result. No cyst lining, however, was evident histologically.

DISCUSSION

This patient presents a tragic problem, since she has had bilateral salpingo-oophorectomies by the age of $10\frac{1}{2}$ years

Submitted, revised, September 23, 1988.

From the Departments of Reproductive Medicine and Pathology, University of California, San Diego, University Medical Center, San Diego, California. Requests for reprints should be addressed to Dr. Paul Wolf, Department of Pathology H-720-A, University Medical Center, 225 Dickinson St, San Diego, CA 92103-1990.

for benign disease. She has now a surgical castration and not only will she be infertile, but without adequate estrogen replacement, she will develop all of the signs and symptoms of premature menopause.

When this unusual condition occurs the first time in young patients, physicians should be aware of the possibility of its recurrence and consider fixation of the other mesosalpinx. Plication may anchor a mobile oviduct and prevent future torsion and infarction. The disadvantage of plication is that, in some cases, it may lead to tubal compromise, and thus to sterility. Possibly an unusually long mesosalpinx is the cause of bilateral torsion, which is a rare event in the absence of ovarian tumors.[2]

The clinical picture simulates acute appendicitis when on the right, infection, ureteral calculi, or colonic disease when on the left, and it may even simulate Mittelschmerz in pubertal girls. These aspects have been adequately described in contributions by Bahary and Eckerling,[3] Sherman and Greenwald,[4] and Hibbard.[5] When the condition is recognized, the treatment is emergency laparotomy; if detected early enough, the adnexa may be salvaged. If infarction has already occurred, removal of the structures is indicated.[6]

The few literature reports on bilateral adnexal torsion cite its occurrence from the age of 2 months to 53 years.[7,8] Ten patients had bilateral torsion simultaneously, while in seven patients it recurred on the opposite side at a later time.[9] Abdominal pain is the most common presenting symptom, followed by nausea, vomiting, and fever, leading to a differential diagnosis of an acute abdomen. Twelve patients developed a palpable abdominal mass, and one half of the specimens demonstrated hemorrhagic infarction of normal structures. In the remaining one half, the specimen disclosed a hydrosalpinx or pyosalpinx.[10] One death has been recorded on the first postoperative day of surgery for torsion with bilateral hematosalpinx. The first patient to be reported with adnexal torsion was that by Sutton in 1890,[11] in a paper on hydrosalpinx. There are well over 300 cases reported with unilateral adnexal torsion.[12,13] The first case report on bilateral torsion is Warnek's description in 1895.[14] The current patient is the 17th so reported. The so-called Kuestner's law,[15] published in 1891, described the twisting of the adnexal pedicle; if the surgeon views the pelvis from the front, then the pedicle on the patient's left side will be rotated to the right in a clockwise direction; the pedicle on the right side will rotate to the left in a counterclockwise direction. In 92 percent of reported cases, the findings have confirmed this law.[16–19]

The following causes of adnexal torsion have been proposed: spasm, mesosalpingeal veins more tortuous and longer than arteries, excessively long oviduct or mesosalpinx, absent mesosalpinx, hydrosalpinx, pyosalpinx, ovarian cysts, ovarian neoplasm, trauma, and following salpingotomy or hysterectomy.[20,21]

Oviduct spasms could possibly result in a twisting effect, as is also possible for engorgement and tortuosity of veins, especially during pregnancy.[22] The same may be true of abnormally long oviducts or absence of a true mesosalpinx. More likely causes are enlarged tubes (hydrosalpinx or pyosalpinx) and especially enlarged ovaries caused by tumors or cysts.[23,24] Trauma and the free end of a postligational tube may conceivably result in twisting.

Torsion and subsequent infarction should always enter the differential diagnostic considerations in acute abdomen of women and female children. Once this diagnosis is established, the surgeon should be mindful of the possibility of recurrence on an as-yet-uninvolved opposite side. He or she may then wish to ascertain possible reasons for such torsion and to undertake preventive fixation or plication of the normal appearing side.

References

1. Anspach BM: The torsion of tubal enlargements with especial reference to pyosalpinx. Am J Obstet Gynecol 1912; 66:513–515
2. Baron C: Torsion of the normal ovary. JAMA 1934; 102:1675–1676
3. Bahary CM, Eckerling B: Torsion of previously normal fallopian tube and ovary in adolescent girls. Gynaecologia 1966; 161:16–20
4. Sherman R, Greenwald JC: Torsion of undiseased fallopian tube in premenarchal female. NY State J Med 1967; 67:1900–1902
5. Hibbard LT: Adnexal torsion. Am J Obstet Gynecol 1985; 152: 456–461
6. Bower RJ, Adkins JC: Surgical ovarian lesions in children. Am Surg 1981; 47:474–478
7. Buss JG: Sequential torsion of the uterine adnexa. Mayo Clin Proc 1987; 62:623–625
8. Chaput M: Contribution a l'étude de la torsion des hematosalpinx complicant les atresies vaginales congenitales. Rev Gyn Chir Abdomin 1906; 10:963–966
9. Dunnihoo DR, Wolff J: Bilateral torsion of the adnexa. Obstet Gynecol 1984; 64(suppl 3):55S–59S
10. Funke A: Stieltorsion bei Hydrosalpinx. Beitr Geburtsh Gynaekol 1904; 7:450–452
11. Sutton JB: Salpingitis and some of its effects. Lancet 1890; 2: 1146–1148
12. Hansen OH: Isolated torsion of the fallopian tube. Acta Obstet Gynecol Scand 1970; 49:3–6
13. Lejars F: Des torsions tubaires. Compt rend Soc d'Obst Gynecol Paed (Paris) 1909; 11:342–345
14. Warnek L: Trois cas de tumeurs des trompes compliquées de la torsion du pedicle. N Arch d'Obstet Gynecol (Paris) 1895; 10:81–82
15. Kuestner O: Das Gesetzmalige in der Torsionsspirale torquierter Ovarialtumorstiele. Zbl Gynaekol 1891; 15:209–210
16. Michel G: Une observation de torsion bilaterale des deux trompes. Ann Gynecol d'Obst 1907; 24:220–221
17. Pozzi MS: Note sur quatre nouveaux cas de la trompe kystique. Compt Rend Soc d'Obstet Gynecol Paed (Paris) 1900; 2:95–97
18. Provost RW: Torsion of the normal fallopian tube. Obstet Gynecol 1972; 39:80–82
19. Reis E: Spontaneous amputation of both fallopian tubes. Am J Obstet Gynecol 1900; 16:325–329
20. Rogers L: Torsion of fallopian tubes. Br Med J 1925; 1:778–780
21. Storer M: Bilateral torsion of the fallopian tubes. Boston Med Surg J 1906; 154:285–287
22. Woodruff JD, Pauerstein CJ: The Fallopian Tube. Baltimore, Williams & Wilkins, 1969, pp 254–257
23. Woolcombe WL: A remarkable case of double pyosalpinx with torsion of both pedicles. Lancet 1901; 2:1584–1586
24. Youssef AF, Fayad MM, Shafeek MA: Torsion of the fallopian tube. A clinico-pathologic study. Acta Obstet Gynecol Scand 1926; 41:292–309