# Update on the management of ovarian torsion in children and adolescents

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***Background:*** **Ovarian torsion is commonly seen in young girls. Unfortunately it is often misdiagnosed because of its non-specific symptoms and lack of diagnostic modalities. This article focuses on the diagnostic challenge and also the changes in the management of ovarian torsion.**

***Data sources:*** **We reviewed original reports on the management of ovarian torsion in young girls published from 1984 till 2014. A literature search was conducted by electronic scanning of five electronic database: MEDLINE, EMBASE, SCI, SSCI and CINAHL. In addition, relevant papers and review articles were hand-searched. The search was limited to English language and human studies. The search was conducted by combining the textwords "ovarian torsion", "adnexal torsion", "adolescents" and "oophoropexy".**

***Results:*** **There are no specific symptoms that can be identified as a pathognomonic feature of ovarian torsion. Ultrasound is a useful diagnostic tool, but it is not always reliable in absence of an enlarged ovary. Laparoscopic detorsion is recognized as the mainstay of treatment regardless the condition of the ovaries. Reports have shown favorable ovarian function after detorsion. The role of oopheropexy remains controversial.**

***Conclusions:*** **Clinicians should be aware of ovarian torsion in girls presenting with abdominal pain. A timely management in this young population can help preserve their ovaries and fertility.**

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***Key words:*** **adolescents; oophoropexy; ovarian torsion**

Review article

## Introduction

Ovarian torsion as a rare entity in childhood and adolescence[1] is an emergency event requiring prompt surgical management. Its incidence ranges from 2/10 000 to 4.9/100 000.[2,3] It accounts for 2.7% of all causes of abdominal pain in children.[4]

Low abdominal pain, nausea, vomiting, low fever and leukocytosis are the usual presenting features. Because of its non-specific symptoms, ovarian torsion is difficult to be distinguished from other entities including acute appendicitis, ruptured ovarian cyst, renal colic and gastroenteritis. Hence, the diagnosis of ovarian torsion remains a great challenge to pediatric surgeons and adolescent gynecologists. A retrospective study[5] showed that misdiagnosis was more likely to make in premenarchal girls than in menstruating women. And absence of an enlarged ovary in this population contributed to the delay in diagnosis. Oltmann et al[6] reported that about 46% of ovarian torsions in children occurred in normal-appearing ovaries.

A literature search was conducted between 1984 and 2014 to provide the clinical symptoms, diagnostic modalities and treatment of ovarian torsion in this young population.

## Pathophysiology of ovarian torsion

Ovarian torsion is associated with pathological changes of the ovary, but it might occur in the normal ovary.[7,8] The exact underlying mechanism of this entity remains poorly understood. Excess mobility in elongated fallopian tubes and ligaments, tubal spasm and sudden change in the intra-abdominal pressure have been postulated as possible causes.[9,10] Increased hormonal activity in the premenarcheal period or even in the

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perinatal period may be associated with the higher risk of ovarian torsion.[10-12]

Ovarian torsion occurs often at the right side of the ovary in a ratio of 3:2.[13] It has been postulated that the presence of the sigmoid colon in the left iliac fossa helps to reduce the motility of the tubal structure, while reducing the risk of left adnexal torsion. However, asynchronous bilateral lesions have also been reported.[7,14]

Mature cystic teratoma which is frequently found to be associated with torsion is followed by corpus luteal cyst and follicular cyst.[10,13] But malignant ovarian tumor associated with ovarian torsion is rare. A review of 424 cases revealed that the incidence of malignancy was only 1.8%.[15] Inflammation and fibrotic changes in malignancy have been postulated to cause adhesion, making the tumor less likely to be twisted.[16]

## Implications of ovarian torsion

Similar to torsion of other structures, venous circulation of the ovary will be compromised followed by arterial circulation. If untreated, this will eventually lead to ischemia and necrosis of the ovary. The absence of unilateral ovaries and tubes in women has been investigated for subfertility. These women have never undergone any operative procedures. Undiagnosed ovarian torsion has been speculated as the cause of its absence.[17,18]

Fatal complications following ovarian torsion have also been reported. Fitzhugh et al[19] reported a case of a four-month-old infant with ovarian torsion and bowel necrosis that lead to cardiorespiratory arrest and eventually to the death of the infant. Havlik et al[20] also reported a case of sudden death from ovarian torsion and suggested that adnexal torsion should be included as a differential diagnosis in cases of sudden death in infancy.

## Presentation and diagnosis of ovarian torsion

The common manifestations of ovarian torsion in post-menarchal adolescents include recurrent abdominal pain followed by nausea, vomiting and occasionally a palpable abdominal mass.[21] Studies[16,22,23] found right-sided pain in more than 50% of the patients, with vomiting in up to 70%; however, fever was found to be an uncommon association with ovarian torsion (16%-22%).[16,22,23] A recent review of 13 girls aged between 7 months and 18 years found that right lower quadrant pain with no pain radiation or migration was the most commonly reported manifestation.[24]

Diagnosis of ovarian torsion is mainly supported by symptoms and ultrasonographic evidence. However, basic laboratory investigations need to be performed to rule out other causes of acute abdominal or pelvic pain, which include urinalysis, pregnancy tests and full blood count. In suspected ovarian tumors, serum levels of tumor markers such as alpha-fetoprotein (αFP), beta-human chorionic gonadotropin (β-HCG), carbohydrate antigen (CA) 125 and CA19-9 should be determined.[25,26]

Plain abdominal X-rays are less effective in detecting ovarian torsion but help to rule out bowel obstruction. The most useful investigation tool is a pelvic ultrasound scan. Diagnostic sonographic features are ovarian enlargement in contrast to the contralateral ovary, presence of multiple small peripherally placed follicles within the ovary, which reflect displacement caused by edema and the presence of fluid collection in the pouch of douglas.[27] Despite the presence of ovarian torsion, doppler ultrasonography[28-30] was normal in 60% of the cases because of the presence of a dual blood supply to the ovary.

Since ovarian torsion presents with non-specific abdominal pain, adolescents with this condition may undergo a CT scan as the first-line radiological investigation. Ultrasonographically, the enlargement of the ovary may be associated with the presence with an ovarian cyst. MRI has been shown to be useful when ultrasound is inconclusive as it has the advantage of identifying the early stages of ovarian edema and hemorrhagic infarction.[27] Although multiple radiological modalities are available, early ultrasound and doppler evaluation could make a prompt diagnosis with a minimal risk of radiation exposure for the patients.

## Changing trends in the management of ovarian torsion

Ovarian torsion needs surgical intervention. In the past, laparotomy has been the surgical approach for the treatment of ovarian torsion. Over the years, laparoscopy has been popularized as it is more reliable than other clinical and radiological assessments. It has been shown to be safe even in young girls,[31-33] with similar outcomes but less morbidities as laparotomy.[34] Cohen et al[35] carried out a retrospective study comparing laparotomy and laparoscopy for ovarian torsion. The outcomes including post-operative ovarian function and macroscopic appearance during the second look surgery were similar in both groups. However, laparoscopy was superior to laparotomy in terms of shorter hospital stay, fewer febrile morbidities and lesser analgesic requirement post-operation.[35] Thus, laparoscopy can be used as a diagnostic tool and more importantly provides

favorable outcomes for girls with ovarian torsion.

When laparoscopy surgery is done in extremely young patients, precautions should be taken into consideration. Insertion of a primary trocar through the umbilical region in a newborn of less than one month old should be avoided, as umbilical vessels may be patent. It is recommended to use a video-guided primary trocar to avoid vessel and visceral injuries. Smaller size trocar/cannula (size 1.7, 3 and 5 mm) should be used.[36,37] Gasless laparoendoscopic single-site surgery is a safe and reliable alternative to the multiport laparoscopic surgery.[38]

Traditionally, oophorectomy has been advocated for the treatment of ovarian torsion.[39,40] The rationale for such operation in the past was based on the following conditions: 1) The "blue-black" ovaries are non-viable; 2) A mere detorsion would trigger possible thromboembolic phenomenon; 3) There is fear of leaving malignant tissue behind.

Currently, there is a more conservative approach, i.e. detorsion, which is reported to be safe and effective in preserving fertility.[10,41,42] Normal appearance of the ovaries have been documented during the second look surgery after detorsion.[35,43-45] Post-operative sonographic assessment is promising as evidenced by the presence of follicles in more than 88% of the detorted ovaries.[1,35,41,42,46-50] Studies[35,43] reported the long-term follow-up results after detorsion. About 5% of the patients with subfertility problems required in vitro fertilization.[35,43] Oocytes were successfully retrieved from detorted ovaries, suggesting that there are functioning ovarian tissues.[35,43]

A study[51] also advocated additional procedures during detorsion, which include cystectomy, aspiration and bivalving. However, only detorsion is recommended to be performed in the "blue-black" ovaries. Cystectomy should be carried out probably six weeks later if the cyst persists. Technically it would be more difficult to enucleate the cyst from the gangrenous ovarian tissue and secure the hemostasis. Styer and Laufer[51] suggested bivalving for the detorted ovaries to reduce the ovarian intracapsular pressure while increasing arterial perfusion. They reported normal follicular function in 4 of the 5 patients who underwent bivalving.

Currently, the reported incidence of pulmonary embolism is only 0.2% in cases of adnexal torsion. But detorsion does not increase the incidence of thromboembolism.[1,35,47]

Ovarian malignancies constitute 1% of all malignancies in children.[13] And only 1.8% of the twisted ovaries are malignant.[15] Most histopathological studies showed that there are no underlying diseases with a low incidence of malignancy.[7,23,27,52,53] Intra-operatively, the ovaries were found to be enlarged because of edematous and ischemic changes after torsion.

In most cases of torsion, the tumor was found to be at early stage, thus it could be cured with surgical resection alone or even with delayed resection after several weeks. If the tumor is in an advanced stage, it can be diagnosed intra-operatively. Nevertheless, correct clinical judgment should be made by the attending surgeon as the ovary may be distorted because of torsion.[54] In case of macroscopic appearance of the bluish black ovary, it is difficult to differentiate a torsed ovary from a torsed teratoma; hence teratoma is diagnosed only after the second-look surgery. However, in case of constant diagnostic doubt, it may be necessary to prepare an intraoperative frozen section to exclude the torsion of a teratoma.[55]

Several factors are associated with malignancy in children. In those aged 1 to 8 years, the presence of abdominal mass or precocious puberty has the greatest percentage of malignancy.[13,53,56-60] Oltmann et al[61] reported that a mass of more than 8 cm in size with a solid area is more likely malignant. Tumor markers like β-HCG, αFP, and CA125 can be used to detect malignancies. The levels of some of the markers can be elevated according to the subtype of the tumor.[62,63] However, a low level of tumor markers is not an absolute indicator to exclude presence of malignancies. There are cases of ovarian torsions with elevated levels of tumor markers in the presence of a benign tumor. Hence, these findings suggest that cautions should be taken during a radical operation in the presence of elevated levels of tumor markers.[64] No doubt, preoperative risk stratification could assist surgeons in their decision-making for preserving the ovaries.

## Role of oophoropexy

Patients with ovarian torsion of the normal adnexa, treated conservatively with detorsion, are at an increased risk of recurrent torsion of the ipsilateral side and the contralateral adnexa.[65,66] Thus, oophoropexy is the most common procedure for the prevention of recurrence of torsion.[67]

Several techniques of oophoropexy have been reported.[68-71] But there is no unified technique nor any consensus about it. The techniques include suturing the ovary to the pelvic sidewall, usually at the level of the pelvic brim[68-70] or to the back of the uterus[71] or to the uterosacral ligaments.[14] In cases of particularly elongated utero-ovarian ligaments, the techniques used are plication of the utero-ovarian ligaments by either

suturing the proximal and distal ends together[69,72] or by shortening the ligaments with placement of an endoloop (Ethicon, Somerville, NJ).[73] These procedures have been performed in recent years via laparoscopy; but measures must be taken to identify the iliac vessels and ureters before plication.[14,74] It is recommended that permanent, non-absorbable sutures be used for all procedures.[75] Surgical clips have also been used to pex the ovary to the sidewalls.[74]

Oophoropexy, however, is controversial over whether it should be performed, whether it should be done at emergency surgery or later, which method is better, and whether one or both sides should be fixed.[14,75] Oophoropexy has been shown to reduce future fertility because of interference with fallopian tubal blood supply or tubal function or with ovarian communication with the fallopian tubes.[21,64,68,76] Fuchs et al[75] followed up six patients after oophoropexy and found that torsion recurred only in one patient. The six patients resumed spontaneous menstruation and two of them conceived and gave a birth. Another consideration is the risk of an endoloop placement that leads to tissue necrosis.[75]

Plication of the utero-ovarian ligaments is the preferred technique of oophoropexy as it has less effect on future fertility. Besides, it is easy to perform with laparoscopy and appears to have few post-operative complications.[74-76] Although oophoropexy remains controversial in the treatment of primary ovarian torsion, it is feasible in certain clinical situation such as recurrent torsion, loss of contralateral ovary and anatomically vulnerable ovary.

## Conclusions

Ovarian torsion should be suspected in girls with abdominal pain. Its diagnosis remains a great challenge because of its non-specific manifestations and lack of diagnostic tools. Laparoscopic detorsion is the treatment of choice regardless the color of the ovaries during the surgery. Oophoropexy should be individualized in patients with ovarian torsion.

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