Adnexal torsion in adolescents: update and review of the literature

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Purpose of review
Adnexal torsion is a surgical emergency, which requires immediate recognition and prompt treatment. This article aims to systematically illustrate the most relevant approach to follow in cases of suspected adnexal torsion in adolescents. We based our evaluation of the role of clinical features, laboratory tests, and imaging on recent literature, till we consider the most appropriate surgical treatment.

Recent findings
The literature describes the conservative treatment for adnexal torsion, consisting of detorsion, as the best surgical approach to guarantee the future reproductive capacity of patients. Recent experimental studies have focused on the consequences of detorsion, which may cause ischemia-reperfusion damage and have speculated about the role of different drugs for its avoidance.

Summary
A systematic review of physiopathology, clinical picture, and imaging of adnexal torsion is certainly useful to identify as soon as possible this emergency condition. An early identification and an adequate treatment are indispensable in order to achieve a proper outcome for the patient. The choice of the appropriate surgical approach can be challenging and should be made on the basis of an accurate evaluation of the ovarian lesion. Therefore, it would be useful to perform an intraoperative frozen section analysis in selected cases.

Keywords
adnexal torsion, adolescent, ovarian torsion, surgical treatment

INTRODUCTION
Adnexal torsion is a surgical emergency defined as total or partial rotation of the ovary, the fallopian tube, or both around its vascular axis causing impairment of blood flow to adnexa. The expression ‘isolated tubal torsion’ (ITT) is used when tubal torsion is not associated with torsion of the ovary [1].

A literature review was carried out between 2013 and 2014 (focusing especially on 2014) in order to present the most up-to-date approach to diagnosis and treatment of adnexal torsion.

Adnexal torsion is relatively frequent in women of reproductive age, although it can be seen in any age group, from children to postmenopausal women. Its incidence in women under 20 is estimated at 4.9/100 000, with a mean age at presentation of 12 years [2]. It is reported to be the fifth most common gynecologic emergency encountered, with a prevalence of 2.7% [3,4,5,6]. In comparison, ITT is a rare condition with an incidence of 1 per 1.5 million [1,7]. Torsion is more common on the right because of the hypermobility of the cecum and ileum in contrast with the relatively fixed sigmoid colon on the left. It can also be secondary to a slightly longer mesosalpinx and uteroovarian ligament on the right side [2,8].

Physiopathology
During adnexal torsion compression of ovarian vessels occurs. Venous and lymphatic outflow is compromised first, as the walls of arteries are thicker and more muscular than those of veins and, therefore, more resistant to compression. This impairment produces an ovarian edema, resulting in the characteristic heterogeneous appearance of the ovary.

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ovary following torsion; arterial blood flow is affected next. If left untreated, the ovary becomes ischemic and then necrotic and complications such as pelvic thrombophlebitis, hemorrhage, infection, and peritonitis may occur [9*,10]. Other complications include calcifications and autoamputation of the ovary. Hasdemir et al. [6] describe an adnexal torsion case with calcifications and speculate that they developed because of chronic hypoxia. Autoamputation occurs through a mechanism of chronic adnexal torsion with subsequent devascularization, necrosis, and detachment [2*,11].

Risk factors
Torsion of normal ovaries is more commonly seen in children and adolescents than in adults, as the uterus is relatively small and the uteroovarian ligaments disproportionately long [2*]. Unlike adults, up to 25% of pediatric patients with adnexal torsion may have normal ovaries without malformations [12,13*]. Excess mobility of the adnexa because of an abnormally long fallopian tube, mesosalpinx or mesovarium, adnexal venous congestion as in premenarchal activity, and jarring movement of the body (vigorous exercise, sudden changes in body position, increases in intraabdominal pressure, trauma) are possible mechanisms that have been suggested in torsion of normal adnexa [13*,14*]. When tubal pathologies are not present, it has been speculated that torsion of the tube may be due to abnormal motility observed during early puberty caused by elevated levels of follicle-stimulating hormone [1*].

Adnexal torsion in children and adolescents may occur due to adnexal disorder such as ovarian cysts and tumors as is seen in adults. The most frequently observed anomalies of ovaries are cystic teratomas, follicular or hemorrhagic cysts, paraovarian cysts, cystadenomas or hydrosalpinx [12].

The risk of adnexal torsion increases when the mass is benign (as malignancies are often fixed to adjacent tissues) and its size is 5 cm or greater [9*]. Ovulation induction, ovarian hyperstimulation, polycystic ovarian syndrome, endometriosis, adhesions, paratubal cysts, infections, and congenital anomalies such as agenesis, hypoplasia, or maldevelopment of müllerian structures also increase the risk of torsion [1*,14*,15,16*].

Clinical presentation
The most common symptom in women with adnexal torsion is acute onset of pelvic or abdominal pain, usually isolated to one side [17**]. This pain may be described nonradiating, constant, or intermittent (depending on whether torsion is partial or complete), mild or intense. Its duration can be variable (from days to months) and there may be a history of similar transient episodes of pain, indicating previous partial torsion. Pain lasting more than 10 h before surgery is associated with an increased rate of tissue necrosis [1*] and it is due to occlusion of the vascular pedicle, with subsequent hypoxia [17**]. It can be associated with nausea, vomiting – peritoneal reflexes – flank pain, anorexia, vaginal bleeding, and bowel or bladder abnormalities [2*,8,10]. Adnexal torsion must be differentiated from appendicitis, kidney stone, mesenteric lymphadenitis, gastroenteritis, corpus luteum hemorrhage/rupture, pelvic inflammatory disease, and necrosis of leiomyoma ectopic pregnancy [9*,12,17**]. Examination findings are often nonspecific, they include normal temperature to low-grade fever, tachycardia, and elevated blood pressure if severe pain is present but these features are more likely with late presentation [2*,18]. To date, no specific laboratory examination for establishing a diagnosis of preoperative adnexal torsion has been described although many studies have been achieved. A pregnancy test, a complete blood cell count, and electrolyte values are usually determined. Most laboratory findings are normal, although a slight leukocytosis may be observed [17**]. Occasionally, a sterile pyuria is described [18]. High serum levels of tumor markers such as CA125 and alpha-fetoprotein may be correlated.
with adnexal torsion and they return to normal levels after surgical removal of the mass [5*,19].

**Imaging**

Preoperative diagnosis is difficult due to vague clinical presentation and aspecific imaging findings of adnexal torsion [20].

Pelvic ultrasonography is the imaging study most commonly used to help diagnose adnexal torsion [9*,17*,21]. The most accurate imaging study available for female pediatric patients presenting with acute abdominal symptoms is transabdominal color Doppler ultrasonography [22]. Asymmetric ovarian enlargement associated with an underlying mass is the most common finding associated with torsion [9*,10,12,17*]. The ovary can lie above and medially to its usual location [17*,23]. Other common findings include free pelvic fluid, edematous, and heterogeneous appearance of the ovary, deviation toward the side of torsion and uniform peripheral follicles as they are pushed to the periphery by the ovarian stromal edema [24]. A torsed fallopian tube can appear dilated, edematous, and fluid-filled if a hydrosalpinx or paratubal cyst is present. The ‘beak sign’ that refers to tapered ends of the fallopian tube can also be described [7*].

Twisting of the vascular pedicle can produce a specific feature, the ‘whirlpool sign’ [10,23]. During ITT, the ovary can be normal and a dilated fallopian tube associated with a whirlpool sign at the end-point of the hydrosalpinx are the only specific features that can be described [25*,26]. The absence of vascular flow is highly suspicious for torsion but it does not occur always; there are cases of torsion in which completely obstructed vascular flow is not described or cases wherein a dual blood supply of the ovary is present [9*,17*]. The role of ultrasound and color Doppler has been widely evaluated by several authors such as Naiditch and Barsness [22] and King et al. [27*].

Asymmetric ovarian enlargement due to an adnexal mass on computed tomography (CT) is the most common finding in torsion, but is nonspecific [9*]. Additional signs of torsion on CT include multiple peripheral follicles, decreased adnexal enhancement on the side of the torsion following administration of contrast, a twisted vascular pedicle, free pelvic fluid, inflammatory fat stranding adjacent to the ovary, and uterine deviation toward the side of torsion. A torsed tube appears dilated [9*,28]. Lourenco et al. [9*] demonstrated that the sensitivity and specificity of CT are not significantly different from that of ultrasonography in diagnosing adnexal torsion.

MRI is not commonly employed as a first-line imaging study in suspected torsion, but can be helpful in pregnant patients or as a problem solver in equivocal cases. Findings are similar to those reported for ultrasonography and CT but MRI offers better soft tissue contrast [10,17*]. Restricted diffusion on diffusion-weighted imaging may be a manifestation of adnexal torsion on MRI [9*].

**Treatment**

Although clinical features and imaging may raise the suspicion of adnexal torsion, the final diagnosis is made during exploratory surgery [20]. The best way to manage adnexal torsion is early diagnosis and immediate surgery [5*,12]. Laparoscopy is considered the best diagnostic and therapeutic approach, especially for the pediatric population [3,29*,30].

There are conservative and radical options for adnexal torsion treatment. It is not always easy to determine whether patients should be treated with detorsion or oophorectomy [31*]. Age, future fertility, menopausal status, and evidence of ovarian disease are all factors considered in the management decision [17*]. To date, conservative treatment is the most recommended surgical approach for adnexal torsion [2*,4*,5*,12,21,32,33*]. In the past the risk of missing an underlying malignancy, thromboembolism after detorsion and a belief that a grossly black hemorrhagic adnexa is irreversibly damaged were arguments in favor of oophorectomy [4*,18,32,33*]. Evidence proves that malignant tumors in children are extremely rare; thus, the probability of leaving malignant tumors is minimal. In addition, pelvic adhesions due to early invasion of adjacent tissue in malignancies can prevent the occurrence of adnexal torsion [5*,12]. It was previously thought that untwisting the torsed adnexa could cause showering of vascular emboli, and, thus, most torsion was managed by removing adnexa without untwisting it. This has been proved untrue as the incidence of pulmonary embolism in cases of adnexal torsion is 0.2% [4*,12]. The presence of edema, inflammation, congestion, and ischemia leads to enlargement of the ovary and an aspect called black-bluish. This may confuse decision-making intraoperatively, resulting in a more inopportune use of oophorectomy [34]. It has been demonstrated that a black-bluish ovary, which does not change its color during surgery, is no evidence of necrosis, and recovery is still likely to occur [4*,17*,34]. Most ovaries demonstrate normal follicular development on ultrasonography and normal Doppler flow after only 6 weeks [18]. If, unfortunately, the ovary appears atrophic on
A second look is necessary in ultrasonography follow-up. In view of that, it may be useful to perform an intraoperative frozen section analysis in selected cases to ensure that the viability of ovarian tissue is preserved and to exclude the presence of necrosis, which is the main histopathological feature highlighted. This observation can be helpful in establishing the definitive surgical approach; if the ovary is ischemic but not necrotic, a conservative treatment consisting in untwisting of the adnexa is more adequate, but, if the ovary is proved to be necrotic, its removal becomes appropriate.

In case of benign neoplasms and cystic lesions, a detorsion associated with synchronous tumorectomy or cystectomy can be performed, but if this is not possible because of the edema and the structural abnormalities of the ovary, surveillance is suitable in order to observe the evolution of the lesion and subsequently perform a second look laparoscopy avoiding oophorectomy. When a suspected malignancy is confirmed by biopsy, an oophorectomy is required. Figure 1 shows the different surgical approaches for adnexal torsion. Neosalpingostomy in premenarchal girls with ITT has been reported as a treatment option that preserves the fallopian tube.

Oophoropexy is defined as a surgical technique aimed at limiting ovarian mobility and preventing further adnexal torsion. The role of oophoropexy is still debated. Kurtoglu et al. affirm that it must be considered at the time of first oophorectomy to prevent torsion of the remaining enlarged ovary; furthermore, the authors recommend a contralateral ovarian fixation in addition to detorsion and pexy of the affected ovary. Sheizaf et al. suggest that even in high-risk cases, the primary management should not include oophoropexy, which may have irreversible consequences and does not necessarily prevent recurrence.

**Effects of reperfusion after detorsion: experimental studies**

Adnexal torsion produces ischemic damage. The treatment goal is to restore blood flow and improve tissue perfusion in ischemia through detorsion. During reperfusion, tissue damage can be more severe than during ischemia because of oxygen-derived radicals (reactive oxygen species) such as hydrogen peroxide, hydroxyl radicals, and nitric oxide, which accumulate after detorsion.
process is known as ‘ischemia–reperfusion injury’ (I/R injury) and it can determine impairment at systemic and tissue levels [38]. Countless studies have investigated whether various substances can prevent I/R injury, which can occur during untwisting of adnexa [36]. Assuming that detorsion can provoke this injury and alone is insufficient to protect ovarian reserve as the follicle count decreases (Ozler et al. [38]), several agents, including antiinflammatories and antioxidant drugs, have been evaluated to determine whether they have a protective effect [36]. Experimental studies were performed on rat ovaries to demonstrate the role of different drugs in the protection of ovarian function [36, 39–53]. Gradual detorsion can also reduce reperfusion injury in a rat model of ovarian torsion [37]. In contrast with these assessments, Yucel et al. [37] and Bozdag et al. [31*] affirm that follicle reserve is not reduced after detorsion and it is not influenced by duration and intensity of ovarian damage.

Outcome

The late effects of radical and conservative surgical approaches on fertility remain uncertain [55]. Studies in adults suggest an association between type of ovarian surgery and subsequent fertility as measured by achievement of pregnancy. Because this is an unrealistic outcome variable in the pediatric population and assuming that menstrual events are considered an appropriate surrogate assessment of gonadal function, Zhai et al. [55] conducted a study including an examination of menstrual events in women who required ovarian surgery. Women who underwent a conservative procedure, in contrast to those who underwent unilateral oophorectomy, showed a notable increase in menstrual irregularity and painful menses, perhaps due to the presence of an altered ovary. Menstrual cycle irregularity and secondary dysmenorrhea are likely associated with reduced fertility and impaired gonadal function. The results of this study suggest that loss of a single ovary does not adversely affect gonadal function, as estimated by effects on menstrual regularity, nor does it appear to compromise future fertility. Even though the authors recommend a conservative approach when possible, the evaluation of these effects throws new light on outcomes after treatment for adnexal torsion. This concern is partially shared by Bellati et al. [56*] who suppose that the removal of one ovary does not significantly worsen the female fertility outcome. On the contrary, the concept that resection of the unilateral affected adnexa as radical treatment can have a negative impact on fertility persists [4***]. Many studies affirm that conservative laparoscopic surgery is the best surgical approach for preserving ovarian function permitting normal development of puberty and maximizing the future reproductive potential of women of reproductive age with adnexal torsion [4***, 5**, 12, 13*, 18, 33**].

CONCLUSION

Adnexal torsion should be considered in female adolescents with acute lower abdominal pain as it can present as an emergency condition. Diagnosis is difficult as clinical features and imaging are nonspecific and a definitive diagnosis is made only during surgery. Surgical treatment should be conservative in order to increase the future reproductive potential of these patients. It may be useful to perform an intraoperative frozen section analysis in selected cases to assess the viability of ovarian tissue and potentially avoid a more aggressive approach. At present, experimental studies investigate the role various substances may play in preventing I/R injury and improving the outcome of these patients.

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Conflicts of interest

There are no conflicts of interest.

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Papers of particular interest, published within the annual period of review, have been highlighted as:

* of special interest
** of outstanding interest


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