The purpose of this study is to discuss the surgical treatment for ovarian torsion in children and adolescents with a focus on the procedures of adnexal conservation surgery and its frequency in the literature of the last 10 years. We retrospectively reviewed the medical charts of 127 operative ovarian lesions including 30 ovarian torsions (23.6%) treated in two pediatric centers over a 10-year period. Age at presentation, presenting symptoms, diagnostic studies, surgical procedure and pathological findings were analyzed. Mean age was 13.7 years. Conservative surgery has been performed in 46.7% of the cases and laparoscopic approach in 40%. Ovarian torsion occurred in 56.7% on ovaries with functional lesion, in 23.3% on normal adnexa and in 20% on ovaries with benign neoplasm. The article includes a literature review (2000–2010) and a statistical analysis which shows a slow increase in conservative surgery from 28 to 45%. Laparoscopic surgery accounts for 23.5%. Literature review shows 40.5% normal adnexa, 33.2% non-neoplastic lesions, 25.3% benign neoplasms and 1% malignant neoplasms. The surgical treatment of children and adolescents presenting adnexal torsion should be practiced as an emergency and it should be more conservative as possible in order to maximize the future reproductive potential.

Keywords: adolescents, children, conservative approach, ovarian disease, ovarian torsion, ovarian surgery

Introduction

Ovarian torsion is the twisting of the ovary on its vascular support. When ovarian torsion remains undiagnosed, blood supply becomes compromised resulting in tissue necrosis. It is important to make an immediate diagnosis and provide treatment in order to save the ovary. Ovarian torsion can occur at all ages, pediatric cases represent the 15% [1], and the estimated incidence is 4.9/100,000 females aged 1–20 years [2]. The twisting of the ovary may involve a normal ovary or an ovary with functional pathology, benign or malignant neoplasm [3,4]. In children torsion on a normal ovary is more frequent because of the greater length of the ovarian pedicle [5–10]. An abnormally long fallopian tube, mesosalpinx or mesoovarium causes an excess of mobility of the adnexa, determining a higher risk of torsion [5,6,11–14]. The clinical presentation of ovarian torsion is unspecific and, at the present time, there is not a reliable method to confirm the diagnosis pre-operatively, sometimes diagnosis is during surgery [5,13]. The resection of the affected adnexa, called as radical treatment, can have a negative impact on fertility, especially in case of recurrence or bilaterality [15], while adnexal conservation surgery, performed as soon as possible, seems to be the best surgical approach in order to maximize the future reproductive potential of the girl. The danger of missing malignant pathology, the risk of thromboembolic complication and the fear that a severe ischemic ovary is non-viable, have led the use of oophorectomy [1,2,14,16–20]. Several studies show a low risk of malignancy and thromboembolism associated with ovarian torsion, and that the macroscopic appearance of ovary is not a true indicator of the degree of ischemia [1,11,21]. These pieces of evidence have suggested the possibility of more conservative surgical treatment.

The purpose of this article is to report our experience on the surgical approach to ovarian torsion in children and adolescents, presenting 30 cases, a literature review of the last 10 years with statistical analysis including 3858 cases.

Materials and methods

This study included all pediatric cases with ovarian masses surgically treated from 2001 to 2010 at our institutions. After the Institutional Review Board approval, the informed consent was obtained from all parents prior to the study. Age at operation and presenting symptoms were analyzed for each patient. Clinical examination, trans-abdominal Ultrasonography (US) and Color Doppler Ultrasonography have been performed in all patients, while selected patients underwent trans-vaginal Ultrasound, standard abdominal x-ray examinations, Computed Tomography (CT), Magnetic Resonance Imaging (MRI) and evaluation of serum tumor markers (CA-125, α-FP, β-hCG and CA-19.9). The nature of the masses was defined by histopathological features using intraoperative frozen section with definitive histological analysis. The histological diagnosis followed the WHO classification [22]. Surgical procedures were led by laparoscopic or open surgery and were performed using either the conservative or the radical approach. Adnexal conservation treatment was practiced as detorsion alone or detorsion followed by subsequent surgical cystectomy or tumorectomy with ovarian tissue preservation and eventually oophoropexy. All children treated conservatively underwent US follow-up at postoperative weeks 1, 4 and 8, then every 3 months up to 1 year.

This article includes a literature review carried out using PubMed and Ovid to research articles published from 2000 to
surgery was 12 h (range: 7 h–4 days). Torsion occurred on the right side in 21 (70%) patients and on the left in 9 (30%) patients.

The surgical approach in children related to 1990 and 2010 respectively. The Spearman's R for the correlation between conservative surgery and radical surgery [35].

Results

Over a 10-year period, 127 surgical interventions for ovarian lesions in 120 children and adolescents have been evaluated (7 presented asynchronous bilateral lesions). Ovarian torsion cases were 30, in 29 children with a mean age of 13.7 years (from 2 months to 18 years). We observed: 61 (48%) functional cysts; 54 (42.5%) benign neoplasms and 5 (4.0%) malignant neoplasms. The ovarian torsion occurs on ovary with hemorrhagic cysts in 10 cases (33.3%), follicular cysts in 7 cases (23.3%), mature cystic teratomas in 5 cases (16.7%) and cystadenoma in one case (3.3%); there were 7 torsions (23.3%) on normal ovaries, including one asynchronous recurrence of a left normal ovary 8 months later the right one (Figure 1). Abdominal pain was the main symptom for ovarian torsion, presenting acute pain in 26 cases (89.6%) and cyclic pain in 3 (19.4%); nausea and vomiting occurred in 17 cases (56.7%) and 6 cases (20%) presented fever. The white blood cell count was elevated 63.3% of the time with a mean count of 12500 cells/mm³. Median delay between initial onset of symptoms and surgery was 12 h (range: 7 h–4 days). Torsion occurred on the right side in 21 (70%) patients and on the left in 9 (30%) patients.

Imaging studies of patients with ovarian torsion included trans-abdominal and color doppler US in all cases, trans-vaginal US in 6 cases (20%), standard abdominal x-ray examinations in 10 cases (33.3%), CT and MRI in 8 cases (26.7%). The sonographic appearance of the lesions was: simple lesions for all follicular cysts and 2 hemorrhagic cysts (20%); complex lesions for 8 hemorrhagic cysts (80%), 3 mature cystic teratomas (60%) and one cystadenoma; solid lesion for 2 mature cystic teratomas (40%). Ovarian enlargement and the presence of small cystic structures around the periphery of the ovary were the sonographic appearance of the ovarian torsion on normal ovaries. The mean diameter of the ovary, measured by ultrasound, was 6.7 cm (from 5.1 to 13 cm). Free fluid in the Pouch of Douglas was present in 8 cases (26.7%).

Reoperative Color Doppler Ultrasonography examination revealed decreased or absent blood flow to the involved ovary in 19 cases (63%) and normal in 11 cases (37%). Because of the clinical emergency, tumor marker levels have been measured in 16 cases, and they were high in one case: α-FP > 12 ng/ml in presence of a mature cystic teratoma complicated by ovarian torsion. This value has normalized 1 month after surgery.

We used a laparoscopic approach in 12 patients (40%) and the open one in 18 (60%). The cases practiced in open were performed mostly in the first period of observation. Adnexal conservation surgery was performed in 14 cases (46.7%) and the surgical procedures were detorsion alone in 5 children (35.7%) with normal ovaries, including 4 ovaries presenting a black-bluish macroscopic appearance at surgery; detorsion followed by cystectomy in 8 children (57.1%) with 4 hemorrhagic cysts and 4 follicular cysts; detorsion followed by tumorectomy in one girl (7.2%) with a mature cystic teratoma.

The resection of the affected adnexa was necessary in 16 children (53.3%) because the ovarian tissue necrosis had progressed. Radical treatment occurred on 9 functional lesions (56.3%), 2 normal ovaries (12.5%), and 5 neoplastic benign lesions (31.2%). Oophoropexy was performed in 4 cases (13.3%). All these torsions occurred on the left ovary, and we practiced oophoropexy by fixing the right ovary to the lateral part of the pelvic wall in 2 cases, and to the posterior wall of the uterus in the other 2 cases.

US follow-up was performed in all children treated by conservative surgery, and in all cases, including black-blush ovaries, we observed recovery of the ovarian tissue with ovarian volume normalization, nearly like the contralateral, in a time between 3 and 6 months.

Our literature review about ovarian torsion considered a total of 3858 pediatric cases with mean age of 14 years. The surgical treatment was radical in 2377 cases (61.6%) and conservative in 1481 cases (38.4%). The ovarian histopathology finding includes 1734 cases. The pediatric torsion occurs on normal ovary in 703 cases (40.5%), on ovary with non-neoplastic lesions in 575 cases (33.2%) and on ovary affected by benign ovarian neoplasms in 438 cases (25.3%). Ovarian torsion in presence of malignancy happened in 18 cases (1%).

The right-left side, the laparoscopic or open approach, and the oophoropexy practice were described for a total of 298 cases and they were respectively: 184 right ovaries (61.7%), 228 cases (76.5%) and 23 oophoropexy practice (7.2%).

The graph in Figure 2 shows the statistical analysis which presents, on the horizontal axis, the median year for the period during which the study was performed and each point, corresponding to the axis of ordinates, is associated with the percentage of work performed by conservative surgery. The median years coinciding with separate studies, are analyzed cumulatively. The size of the circles is directly proportional to the number of cases. It was finally calculated a linear weighted average of the number of cases accumulated for each year median. The graph shows an increasing trend of cases treated with conservative surgery, from 28% to 45% related to 1990 and 2010 respectively. The Spearman's R for the two surgical approaches, conservative and radical, is 0.122 (p < 0.01 one tail) permitting to consider the analysis to be significant.

Discussion

Ovarian torsion accounts for up to 2.7% of all cases with acute abdominal pain in children 18,26. This is the main symptom, presented in 83% to 100% of cases [7,13,31], and it may be associated with other symptoms, such as fever (from 18% to 22%)
Surgery for ovarian torsion

Therefore its clinical presentation is unspecific and it can mimic many abdominopelvic surgical diseases. Unfortunately only about one half of the patients are suspected of adnexal torsion at the time of the first clinical diagnosis, it is often confused with acute appendicitis also for possible presence of leucocytosis (50–82%) [8,21,26,32,36]. Although acute presentation is more frequent, patients ranging from 4% to 28% present a history of cyclic abdominal pain, this is due to torsion and spontaneous detorsion of the ovary [8,11,14,15,31]. During the diagnostic stage, abdominal mass effect and calcification on plain x-ray study suggest the presence of mature cystic teratoma [13,14,16,24], but ultrasonography is the primary diagnostic method employed for the evaluation of suspect ovarian torsion [17,37,38]. The most common sonographic finding of torsion in normal ovaries is ovarian enlargement [7,13,17,37], and the presence of fluid-debris levels within small cystic structures, around periphery of the ovary, is greatly suggestive of torsion of normal ovary, this feature is presented in 13–45% of cases [13,17,36,37].

Color Doppler diagnostic contribution remains controversial because of the dual blood supply to the ovary, thus the presence of vascular flow to the ovary does not exclude torsion moreover, intermittent torsion can lead to normal flow if the adnexa are not twisted at that moment [5,7,14,36,37]. Studies report the absence of Doppler flow in torted ovaries from 38 to 62% [36–38] and Linam et al. [38] in their retrospective case-control study observe the absence of doppler flow also in 1/3 of controls. The size and the weight of ovarian lesions, specially of mature cystic teratoma, are implicated in promoting torsion [1,5]. Oltmann et al. [7] observe 83% of torsed ovaries have a US measurement of 5 cm or larger while Rousseau et al. [13] observed only torsions under 15 cm, because large tumors are quite fixed, enclosed in the pelvis. The same occurs for torsion in a malignant pathology, this is less frequent because the tumor creates inflammation and adhesions to surrounding tissues reducing mobility and consequently torsion [5,17,20]. Therefore, in pediatric age, the risk of malignant ovarian neoplasm associated with torsion ranges from 2% to 6% [7,20,21,32,39]. Because of the acute symptoms, CT and RM are performed in doubtful cases only and their common imaging features for ovarian torsion are enlarged adnexa or evidence for adnexal mass, evidence of vascular congestion, ascites, tubal thickening and uterine deviation to the twisted side [40,41]. In emergency, levels of tumor markers such as β-HCG, α-FP, CA-125, LDH are not routinely analyzed. The literature reports cases of ovarian torsion with increase of tumor marker in presence not only of malignant but also benign neoplasm [42,43]. Savic et al. [20] reported a series of pediatric ovarian torsion and a review of literature [32] with increase of LDH, β-HCG, αFP, CA-125 in 7 malignant germ cell tumors, increase of CA-125 in the presence of 5 benign neoplasms and 3 non-neoplastic lesions. McCarthy et al. [44] reported a case of a 6-year-old girl with ovarian torsion in presence of increased CA-125 where the pathological diagnosis was ovarian necrotic cellular debris. Takeda et al. [25] presented the case of a 17-year-old girl with dysgerminoma complicated by ovarian torsion and increase of LDH and β-HCG. We observed a
case of ovarian benign mature teratoma complicated by torsion in association with increased α-FP. This finding suggests to be cautious in performing a radical surgery in presence of elevated tumor markers.

In the past concern for pulmonary embolism has induced to practice radical surgery but the literature shows only two adult cases of pulmonary embolism, both occurred after salpingo-oophorectomy without detorsion [19,45]. Whereas, in pediatric age there was only one case, reported in a 16-year-old patient, of the right common iliac vein thrombosed after left ovarian detorsion and cystectomy [46].

In medical literature, the standard procedure for many years has been the removal of the twisted ovary, but our statistical study shows that conservative treatment is slowly increasing from 28% to 45% in the last 20 years. This is an important finding because it reflects the greater interest in conservative surgery.

In ovarian torsion, laparoscopy is the recommended surgical approach if technically possible. It provides the benefit of first intervention when the diagnosis is unclear, decreasing the diagnostic delay and increasing conservative surgery [5,7,17,27,47]. Laparoscopic approach has an overall figure of 23.5% in the review of the literature. With reference to Garliner et al. [17] when the twisted ovary is below 75 mm, the surgical approach should be laparoscopy, given the usual benign nature of the tumor under this size, contrariwise, laparotomy should be performed.

The presence of edema, inflammation, congestion and ischemia determines an enlargement in dimensions of the ovary and an aspect called black-bluish. This aspect may confuse decision making intraoperatively, resulting in a more aggressive use of oophorectomy [21,48]. Most recent studies have shown that there is no valid clinical method at hand predicting the viability of the ovary and that the black-bluish macroscopic appearance of the ovary is not a true indication of the degree of ischemia [1,11,14,16,21]. Taskin et al. [49] in their study analyzed a rodent model of ovarian torsion and performing histological analysis of all black-bluish ovary they have concluded that adnexal integrity is not correlated with gross ischemic appearance. To obtain information on the viability of ovarian tissue and to exclude the presence of necrosis, it would be useful perform an intraoperative frozen section analysis. Moreover, it is demonstrated that a black-bluish ovary, which does not change its color during operation, is no evidence for a necrosis, and recovery is still likely to occur [1,2,5,11,16–18,21]. In particular Celik et al. [11] reached 92% of adnexal functional integrity, shown by ultrasonography follicle growth. To decrease ovarian intracapsular pressure and facilitate ovarian reperfusion and recovery, after untwisting the ovary, some authors proposed ovarian bivalving [13,48], whereas others do not agree on this procedure, because it could appear excessive on an ovary already insulted by torsion [17]. Svensson et al. [9] describe additional hyperbaric oxygen therapy which increases the tissue oxygen tension, increases perfusion in injured tissue and improves function of leukocytes with general anti-inflammatory effects. However, on previously normal ovaries complicated by torsion, conservative management consists in detorsion alone, even if they present severe ischemic-hemorrhages, without no necrosis yet. After conservative treatment a clinical and a sonographic follow-up permit to observe two possible evolutions: the ovary can retrieve its functional integrity by follicular presence, with back to normal size or not, or, unfortunately, it can become atrophic [11,14,16]. In their study, Wang et al. [39] observe an ovarian recovery with normal size and normal follicular development in 33/35 (94.3%) cases treated conservatively while 2/35 cases (5.7%) become atrophic.

When cystic masses are diagnosed, detorsion of the adnexa with synchronous cystectomy can be performed [1,17,33]. Otherwise it is possible to realize detorsion alone with surveillance in order to observe the evolution of the lesion and in case of its persistence or enlargement in size practice a second look [5].

In presence of a solid or heterogeneous mass it is necessary, after detorsion, to define if there is an underlying neoplasm [7] and for this reason some authors suggest the use of intraoperative frozen section [39,50]. When a malignant lesion is diagnosed, staging is realized and an appropriate surgical procedure can be adopted [5]. In case of benign lesions synchronous tumorectomy can be performed but this procedure is technically difficult and it may cause further insult to the remaining ovarian tissue with partial or complete involution of the ovary [17,18,33]. Therefore it can be useful to practice detorsion alone with subsequent surveillance. Different authors [11,14,16–18,21,33,51] suggest re-examining these patients from 1 to 8 weeks after the acute episode, using ultrasonography and tumor marker levels because the edema and inflammation typically resolve and, at that time, ovarian tumorectomy can be performed in a second look [13].

As to oophoropexy, this practice is proposed in order to prevent recurrence of ipsilateral or contralateral adnexal torsion. Studies show a risk of asynchronous bilateral ovarian torsion from 5% to 13% [13–15,17]. This risk is higher if the first episode occurs on the left side and on an ovary without any pathology [14]. In fact, the left ovary has a lower frequency of torsion because of the presence of the sigmoid colon which limits mobility [5–8,13,37]. In our review of literature we observed a frequency of 7.2%. Oophoropexy is achieved using a non-resorbable swaged suture and by fixing the ovary to the lateral part of the pelvic wall; to the posterior wall of the uterus; to the posterior peritoneum just below the bifurcation of the common iliac vessels [5,17]. Otherwise it is possible to use the procedure of uterus ovarian ligament plication which provides the means for shortening the uterus-ovarian ligament and thus reduces ovarian laxity [5,15]. Possible risks of this procedure may include adherence and disturbance of the anatomic tubo-ovarian relationship with displacement of the ovary from the fallopian tube and subsequent infertility [12,15,33]. Because no randomized, double-blind prospective studies, exist to solve the question of the success of the pexy and its impact on future fertility, the use of oophoropexy is controversial and not generally recommended for routine use [7,16]. Therefore, only selected patients may benefit from pexy: those undergoing pelvic radiation for the treatment of malignancy, those presenting bilateral torsion or recurrent ipsilateral torsion and when torsion occurs on an ovary with a malformed or excessively long utero-ovarian ligament [5,7,16].

In conclusion, the conservative surgical treatment of ovarian torsion in children and adolescents, as evidenced in our study, requires a specific knowledge of the physiopathology as well as clinical and diagnostic aspects of the condition. In order to maximize ovarian salvage it is necessary to perform diagnosis and therapy as soon as possible. The laparoscopic approach practiced in emergency, allows to reduce diagnostic delays. The correct surgical procedure should be decided after ovarian detorsion and only after intraoperative histopathological characterization of the lesion, especially in presence of solid or mixed imaging.

**Declaration of Interest**: The authors report no declaration of interest.