

Asynchronous bilateral ovarian torsions in girls-systematic review

Maja Raicevic, Amulya Kumar Saxena

London, United Kingdom

Background: Bilateral ovarian torsions with complete loss of ovaries is devastating. This study analyzed the literature on bilateral ovarian torsions in girls to evaluate surgical options and outcomes.

Methods: Literature was searched on Pubmed® (1987-2014) using terms "bilateral", "adnexal", "ovary", "torsion" and "children". Data were collected on age, surgical preference, pathology and outcomes.

Results: Thirteen articles were identified, and 9 met the inclusion criteria (5 case reports, 4 original articles); and analyzed 17 girls (mean age: 8.75 years, range: 1-16). Bilateral oophorectomies ($n=4$), ipsilateral oophorectomy of severely affected ovary and contralateral oophoropexy ($n=10$), and detorsion of bilateral ovaries and bilateral oophoropexy ($n=3$) were performed. One torsion recurrence occurred after two oophoropexies. Laparoscopy and open surgery was done in 2 and 15 girls, respectively. Considering etiology, there were simple tubo-ovarian torsions ($n=8$), polycystic ovary ($n=1$), polycystic ovary associated with Down syndrome ($n=1$) and corpus luteum cyst ($n=1$). No tumors were reported. Serial ultrasound follow-ups of ipsilateral oophorectomy and contralateral oophoropexy ($n=5$) confirmed follicular function ($n=4$) and viability and position of the ovary ($n=1$).

Conclusions: Though extremely rare, school age girls present bilateral ovarian torsion. Ipsilateral oophorectomy and contralateral detorsion with oophoropexy has been the preferred approach.

World J Pediatr 2017;13(5):416-420

Author Affiliations: Department of Pediatric Surgery, Chelsea Children's Hospital, Chelsea and Westminster NHS Fdn Trust, Imperial College London, United Kingdom (Saxena AK); Clinic for Pediatric Surgery and Orthopedic Nis, Clinical Center Nis, Serbia (Raicevic M)

Corresponding Author: Amulya Kumar Saxena, MD, PhD, DSc (hon), FRCS (Glasg), Consultant Pediatric Surgeon, Department of Pediatric Surgery, Chelsea Children's Hospital, Chelsea and Westminster Hospital NHS Foundation Trust, Imperial College London, 369 Fulham Road, London SW109NH, United Kingdom (Tel/Fax: +44-20-3315-8885; Email: amulya.saxena@nhs.net)

doi: 10.1007/s12519-017-0052-3

Online First June 2017

©Children's Hospital, Zhejiang University School of Medicine, China and Springer-Verlag Berlin Heidelberg 2017. All rights reserved.

Key words: asynchronous; bilateral; outcomes; ovarian torsion; surgery

Introduction

Ovarian torsion is the most frequent gynecological emergency in children.^[1] Although the adnexal torsion is most frequently unilateral, cases of bilateral synchronous or asynchronous ovarian torsion have been reported. Ovarian torsion was described for the first time by Sutton in 1890, and the first case of bilateral ovarian torsion reported was in 1895 by Warnek; however bilateral asynchronous ovarian torsion in childhood was first described by Baron in 1934.^[2,3]

Asynchronous bilateral ovarian torsion (ABOT) is defined as torsion of each ovary at different points in time.^[3] The presenting symptoms of ovarian torsion are often non-specific and may include acute onset of lower abdominal pain, nausea, and vomiting.^[4] Clinical imaging and presentation do not offer definitive diagnosis but rather indications of possible torsion which has to be evaluated in the context of differentials.^[5] When considering the differentials, appendicitis has been found to be the most common preoperative diagnosis.^[6] Functional ovarian cysts are very common during peri-pubertal years, may be also an additive predisposing factor of adnexal twisting.^[7]

Factors contributing towards ABOT are still unknown. Higher recurrence rate has been noticed in patients with previously normal adnexa, especially in prepubertal girls.^[3,4] The main factor that makes this entity stand out is that complete loss of ovarian tissue can be devastating for patients and their families. The loss of ovarian tissue can be a result from delay to surgery, from unrecognized fear of untwisting the adnexa, or failure to protect any residual ovary (ipsilateral and contralateral) from subsequent torsion.^[3] The loss of ovarian tissue is an extremely rare event, and there are no guidelines or consent statements with regards to surgical approach to ABOT. This study performed a literature based analysis on ABOT in girls and evaluated their outcomes to analyze the evidence in management.

Methods

The literature was searched on Pubmed® (1987-2014) using terms "bilateral", "adnexal", "ovary", "torsion" and "children". Data was collected with regards to the primary end points of this analysis, age at presentation in the pediatric age group 0-16 years, type of surgery approach, investigations, possible pathology that led to the torsion and follow-up outcomes. Article that did not meet the inclusion criteria of the analysis were excluded from this study.

Results

The 27-year literature search revealed a total of 13 articles on ABOT of which only 9 met the inclusion criteria. These included 5 case reports and 4 original articles; and these articles reported 17 girls with a mean age of presentation of 8.75 years (range: 1-16) that were managed for ABOT. All patients presented with lower abdominal pain, nausea and vomiting. Two girls were febrile with temperatures up to 37.8°C^[2,6] and 1 girl presented with a palpable pelvic mass (although operative finding reported a twisted, edematous ovary with no cyst or tumor).^[8] Except one, all girls underwent ultrasound examination and only in 2 girls no evidence of blood flow on Doppler imaging were found.^[7,8] Erect abdominal films were performed in girls and the findings were reported to be normal.^[2,6] Computed tomography was performed in only 1 girl.^[4] White blood cell counts increased in three girls.^[2,6]

With regards to surgical management, bilateral oophorectomy ($n=4$, 23.5%), ipsilateral oophorectomy of severe affected ovary and contralateral oophoropexy of the "less" affected ovary ($n=10$, 59%), and salvage surgery with detorsion of bilateral ovaries and bilateral oophoropexy ($n=3$, 17.5%) were performed (Fig. 1). There was 1 torsion recurrence after oophoropexies on two occasions in a prepubertal girl; both ovarian ligaments in this case were described as extremely long and ovaries were polycystic.^[4] With regards to surgical

access, laparoscopy was performed only in two patients while open surgical approach was the preferred option in the rest of the 15 girls. The laparoscopic procedures did not report any conversions.

Considering the etiology, the data evaluated found simple tubo-ovarian torsions ($n=8$), polycystic ovary ($n=1$), polycystic ovary associated with Down syndrome ($n=1$)^[1] and corpus luteum cyst in 5.5×4.8 cm diameter ($n=1$). No tumors were reported. Hormonal replacement therapy was administered in one patient who had a right and then subsequently left salpingo-oophorectomy.^[2] No major complications or mortality were found in girls with ABOT.

The level of evidence is poor with regards to the follow-up studies and investigations. Serial ultrasound follow-up investigations were performed in 5 girls who presented with ipsilateral oophorectomy and contralateral oophoropexy. The follow-up period was from 6 months up to 3 years. The ultrasound investigations confirmed follicular function ($n=4$),^[3,5,7,8] and viability and position of the ovary ($n=1$) was confirmed by second look operation after uncertain repeated ultrasound examinations.^[6] Late outcomes were not evaluated in all studies.

Discussion

Ovarian torsion is not an uncommon emergency in girls. The mechanism of ovarian torsion is not known with certainty and some theories explain the possibility of these occurrences. 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 are possible mechanisms that have been implicated in torsion of normal (uterine) adnexa. The presence of ovarian torsion requires prompt diagnosis and urgent surgical intervention to prevent loss of ovarian tissue resulting from ischemia and necrosis.

Recurrent torsion is common in patients whose first torsion episode occurred in the premenarchal period and involved otherwise normal adnexa. Oophoropexy procedures may be considered in patients at risk for torsion recurrence.^[9] However, the state of the ovary and the decision of the surgeon determines if an oophorectomy is performed or a salvaged ovary is left with or without fixation once the torsion is released. In a national survey in the United States, 2041 patients with ovarian torsion were evaluated, of which 1598 (78%) underwent oophorectomy, 126 (6%) oophoropexy, and 317 (15%) release of torsion. Release of torsion significantly increased from 1998 to 2011 (9% vs. 25%; $P<0.05$); also at nonteaching hospitals, there were higher rates of oophorectomy (89.3% vs. 79.5%; $P<0.05$) and lower rates of release

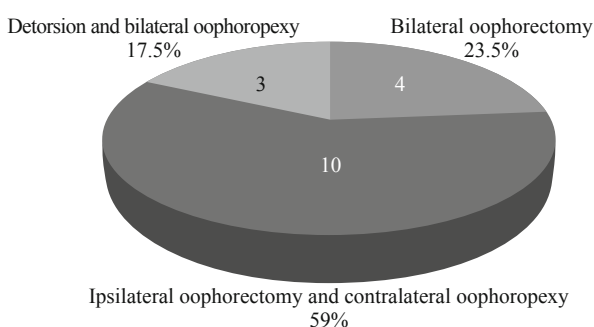


Fig. 1. Cumulative surgical techniques preferred in the last 3 decades in the management of asynchronous bilateral ovarian torsions (pie-chart). However, contemporary approach strongly advocates salvage of ovarian tissue.

of torsion (10.7% vs. 20.5%; $P < 0.05$) compared with teaching hospitals.^[10] Hence, it is difficult to evaluate the rationale behind the treatment in more complex or bilateral manifestations such as ABOT in context of the hospitals where these treatments were performed, experience of the surgical team in terms of managing ovarian pathologies as well as the eras in which the ABOT studies were reported.

Ovarian torsion in premenarchal girls is associated with nonspecific signs and symptoms, and abdominal ultrasound and Doppler imaging may assist in the diagnosis.^[11] The diagnosis does not offer insights into the state of the ovary, which can be determined only during the surgery. In a recent study on 29 females (mean age: 10.3 ± 4.9 years) who underwent surgery for ovarian torsion with detorsion and ovarian preservation in "dusky/purple" ovaries in 21 cases, "normal" in 1, "necrotic" in 1, and not described in 6; all pubertal patients resumed menstrual function and no patients required reoperation for removal of the salvaged ovary.^[12] There also were no instances of postoperative fever or concern for ovarian venous thrombosis. In these patients, average follow-up ultrasonography was performed in 8.1 ± 6.7 months, with 28 patients showing ovarian follicles on the affected side (mean 4.6 ± 1.9 and 4.7 ± 3.3 follicles on the right and left ovary, respectively) and no correlation was found between the sides affected, gross appearance of the torsed ovary or the number of follicles found on follow-up ultrasonography. Hence, long-term analysis of the treatment of ovarian torsion revealed that ovaries treated by detorsion and left in the abdominal cavity preserved their normal anatomy and function. Ovary sparing surgical treatment proved to be safe.^[13] Hence, even if the diagnosis is missed and the ovary is ischemic, detorsion and salvage is recommended.

Diagnosis of ovarian torsion based on clinical or laboratory features that would identify cases of ovarian torsion among girls admitted with abdominal pain could not be identified in a study published in 2013.^[14] However in the same year another report showed that presence of intermittent pain, non-radiating pain and increased adnexal size have been found to be significantly associated with ovarian torsion and it has been suggested that these three parameters are excellent in discriminating ovarian torsion and other causes of acute abdominal pain.^[15] Although, ultrasound scan remains the most useful investigation, with color Doppler ultrasonography able to detect the status of adnexal circulation; however presence of normal flow does not exclude torsion.^[7] Normal Doppler ultrasound findings in a torqued ovary can be one of the reasons for delay in surgery. So, clinical judgement needs to be correlated to the ultrasound studies. Acute lower

abdominal pain in a young female patient, especially one who has had previous similar episodes, should alert the physician to the possible presence of adnexal torsion.

^[16] Recurrent torsion is easier to diagnose owing to the patients' familiarity with symptoms and higher suspicion index.^[4] Worth mentioning at this point would be the role of biomarkers in the investigation. It was reported that interleukin-6 was significantly elevated in surgical patients compared to nonsurgical patients; and CD64 was significantly elevated in appendicitis patients compared to ovarian torsion patients.^[17] However, it must be noted that such investigations may not be available at all pediatric surgical centers.

ABOT presents a special sub-group of patients and this study had accumulated a maximum of 17 reported cases for this analysis. The trend in the literature spans a time of almost 3 decades and the cumulative approach has pointed towards favoring ipsilateral oophorectomy and contralateral detorsion with oophoropexy as the preferred approach in 59% patients. Although a high percentage of patients (23.5%) underwent bilateral oophorectomy, this trend would not be evident with the emerging data which showing evidence of function in salvaged ovaries.^[18] Detorsion and oophoropexy of affected ovary is recommended and in case of bilateral torsions, bilateral oophoropexies should be performed irrespective of the state of the ovarian tissue at the time of surgery.

The use of laparotomy versus laparoscopy is at the discretion of the surgeon but there is agreement in the need for rapid intervention if torsion is suspected.^[6] However, in the present era, more urgent intervention should be considered in girls with lower abdominal pain if ovarian torsion is suspected, with liberal use of diagnostic laparoscopy and without reliance on a definitive diagnosis by imaging.^[19] Explorative laparoscopy is minimally invasive and if a torsion is evident, this mode is therapeutic in detorsion of the ovary and oophoropexy. There are always some concerns in leaving behind "necrotic" tissue in cases of ovaries that have shown severe signs of vascular insults with regards to the post-operative complications. The most common complication of conservative management (salvage of twisted ovaries after severe vascular insult) is postoperative fever,^[6,7] and should be considered in the postoperative management protocol.

Tumors are not associated in asynchronous bilateral torsions according to our analysis, although they add a new dimension to approach.^[20] The presence of tumors must not be overlooked in the management of ABOT. The pitfall in the management of ovarian torsion is to rupture a twisted ovary which has a malignant tumor and to upgrade the tumor due to spillage of its contents.

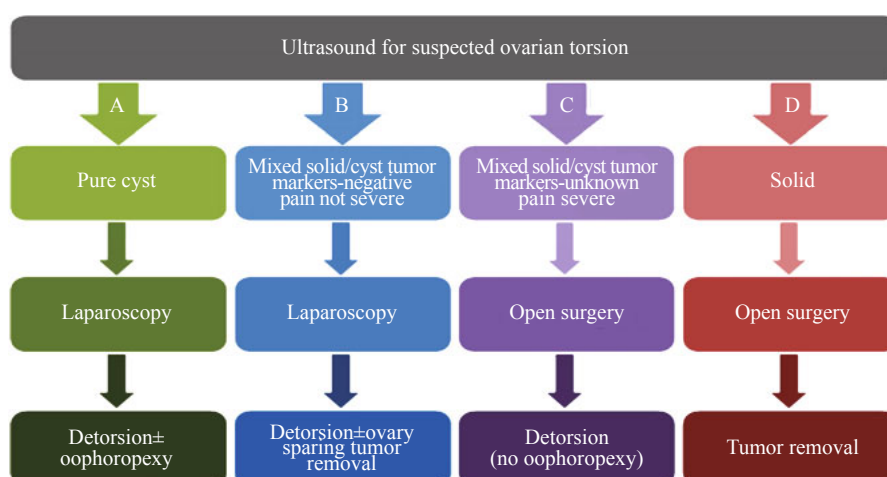


Fig. 2. Proposed algorithm in the management of ovarian torsion, while considering future possibility of asynchronous bilateral ovarian torsion and taking into account the possibilities of tumor presence.

Patients with ovarian torsions present this dilemma in the early management.

An algorithmic approach to this could be as follows (Fig. 2): 1) If ultrasound shows only cystic content, proceed with explorative laparoscopy; 2) If ultrasound shows mixed solid cystic content and the pain is not severe, investigate the tumor makers, if negative proceed with laparoscopic surgery; 3) If ultrasound shows mixed solid cystic content and the pain is not severe, investigate the tumor makers, if positive proceed with open surgery; 4) If ultrasound shows mixed solid cystic content and the pain is severe, and tumor markers cannot be investigated as an emergency, proceed with open surgery and relieve the torsion-but avoid oophorectomy to prevent spillage and upgrading.

In conclusion, the present analysis strongly suggests towards preservation of ovarian tissue, irrespective of the condition of the ovary at the time of surgery, as there is good evidence that these salvaged ovaries demonstrate function. The most common morbidity in conservative management of twisted ovaries is postoperative fever which should be considered in the postoperative management. Girls with ovarian torsion need prompt assessment and surgical assessment preferably with the laparoscopic approach. ABOT presents this sub-set of patients that further demonstrate the need for ovarian preservation during initial surgery, as total loss of ovaries can have devastating consequences for the patient.

Funding: None.

Ethical approval: Not needed.

Competing interest: No conflicts of interests and no relevant disclosures.

Contributors: Raicevic M contributed to the data collection, study

conduct and manuscript preparation. Saxena AK contributed to the study design and conduct, manuscript preparation and final draft. All authors approved the final version of the manuscript.

References

- 1 Khemakhem R, Barhoumi A, Matoussi N, Hidouri S, Ghorbel S, Noura F, et al. The adnexal torsion in children. *Tunis Med* 2008; 86:468-472. [In French]
- 2 Varras M, Akrivis C, Demou A, Antoniou N. Asynchronous bilateral adnexal torsion in a 13-year-old adolescent: our experience of a rare case with review of the literature. *J Adolesc Health* 2005;37:244-247.
- 3 Beaunoyer M, Chapdelaine J, Bouchard S, Ouimet A. Asynchronous bilateral ovarian torsion. *J Pediatr Surg* 2004; 39:746-749.
- 4 Sheizaf B, Ohana E, Weintraub AY. "Habitual adnexal torsions"-recurrence after two oophorectomies in a prepubertal girl: a case report and review of the literature. *J Pediatr Adolesc Gynecol* 2013;26:e81-e84.
- 5 Rousseau V, Massicot R, Darwish AA, Sauvat F, Emond S, Thibaud E, et al. Emergency management and conservative surgery of ovarian torsion in children: a report of 40 cases. *J Pediatr Adolesc Gynecol* 2008;21:201-206.
- 6 Eckler K, Laufer MR, Perlman SE. Conservative management of bilateral asynchronous adnexal torsion with necrosis in a prepubescent girl. *J Pediatr Surg* 2000;35:1248-1251.
- 7 Kurtoglu E, Kokcu A, Danaci M. Asynchronous bilateral ovarian torsion. A case report and mini review. *J Pediatr Adolesc Gynecol* 2014;27:122-124.
- 8 Ozcan C, Celik A, Ozok G, Erdener A, Balik E. Adnexal torsion in children may have a catastrophic sequel: asynchronous bilateral torsion. *J Pediatr Surg* 2002;37:1617-1620.
- 9 Smorgick N, Melcer Y, Sarig-Meth T, Maymon R, Vaknin Z, Pansky M. High risk of recurrent torsion in premenarchal girls with torsion of normal adnexa. *Fertil Steril* 2016;105:1561-1565.e3.
- 10 Sola R, Wormer BA, Walters AL, Heniford BT, Schulman AM. National trends in the surgical treatment of ovarian torsion in children: an analysis of 2041 pediatric patients utilizing the

- nationwide inpatient sample. *Am Surg* 2015;81:844-848.
- 11 Ashwal E, Krissi H, Hiersch L, Less S, Eitan R, Peled Y. Presentation, diagnosis, and treatment of ovarian torsion in premenarchal girls. *J Pediatr Adolesc Gynecol* 2015;28:526-529.
- 12 Santos XM, Cass DL, Dietrich JE. Outcome following detorsion of torsed adnexa in children. *J Pediatr Adolesc Gynecol* 2015;28:136-138.
- 13 Geimanaite L, Trainavicius K. Ovarian torsion in children: management and outcomes. *J Pediatr Surg* 2013;48:1946-1953.
- 14 McCloskey K, Grover S, Vuillermin P, Babl FE. Ovarian torsion among girls presenting with abdominal pain: a retrospective cohort study. *Emerg Med J* 2013;30:e11.
- 15 Appelbaum H, Abraham C, Choi-Rosen J, Ackerman M. Key clinical predictors in the early diagnosis of adnexal torsion in children. *J Pediatr Adolesc Gynecol* 2013;26:167-170.
- 16 Buss JG, Lee RA. Sequential torsion of the uterine adnexa. *Mayo Clin Proc* 1987;62:623-625.
- 17 Reed JL, Strait RT, Kachelmeyer AM, Byczkowski TL, Ho ML, Huppert JS. Biomarkers to distinguish surgical etiologies in females with lower quadrant abdominal pain. *Acad Emerg Med* 2011;18:686-691.
- 18 Dolgin SE, Lublin M, Shlasko E. Maximizing ovarian salvage when treating idiopathic adnexal torsion. *J Pediatr Surg* 2000;35:624-626.
- 19 Piper HG, Oltmann SC, Xu L, Adusumilli S, Fischer AC. Ovarian torsion: diagnosis of inclusion mandates earlier intervention. *J Pediatr Surg* 2012;47:2071-2076.
- 20 Ruttenstock EM, Saxena AK, Schwinger W, Sorantin E, Hoellwarth ME. Pediatric ovarian tumors-dilemmas in diagnosis and management. *Eur J Pediatr Surg* 2010;20:116-120.

Received June 11, 2016

Accepted after revision December 9, 2016