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Ovarian torsion in pediatric and adolescent patients: A systematic review

Roshni Dasgupta ^{a,*}, Elizabeth Renaud ^b, Adam B. Goldin ^c, Robert Baird ^d, Danielle B. Cameron ^e, Meghan A. Arnold ^f, Karen A. Diefenbach ^g, Ankush Gosain ^h, Julia Grabowski ⁱ, Yigit S. Guner ^j, Tim Jancelewicz ^h, Akemi Kawaguchi ^k, Dave R. Lal ^l, Tolulope A Oyetunji ^m, Robert L. Ricca ⁿ, Julia Shelton ^o, Stig Somme ^p, Regan F Williams ^h, Cynthia D. Downard ^q

^a Division of Pediatric General and Thoracic Surgery, Cincinnati Children's Medical Center, Cincinnati, OH

^b Department of Surgery, Division of Pediatric Surgery Albany Medical College, Albany, NY

^c Division of Pediatric General and Thoracic Surgery, Seattle Children's Hospital, Seattle, WA

^d Division of Pediatric General and Thoracic Surgery, Montreal Children's Hospital, Montreal, QC

^e Department of Surgery, Boston Children's Hospital, Boston, MA

^f Section of Pediatric Surgery, CS Mott Children's Hospital, Ann Arbor, MI

^g Department of Pediatric Surgery, Nationwide Children's Hospital, Columbus, OH

^h Division of Pediatric Surgery, University of Tennessee Health Science Center, Le Bonheur Children's Hospital, Memphis, TN

ⁱ Division of Pediatric Surgery, Ann and Robert H. Lurie Children's Hospital of Chicago, Chicago, IL

^j Division of Pediatric General and Thoracic Surgery Children's Hospital Orange County, University of California Irvine

^k Department of Pediatric Surgery, Mc Govern Medical School at the University of Texas Health Science Center at Houston

^l Division of Pediatric Surgery, Department of Surgery, Medical College of Wisconsin, Milwaukee, WI

^m Department of Surgery, Children's Mercy Hospital, Kansas City, MO

ⁿ Division of Pediatric Surgery, Naval Medical Center, Portsmouth, VA

^o Division of Pediatric Surgery, University of Iowa Stead Family Children's Hospital, Iowa City, IA

^p Division of Pediatric Surgery, Children's Hospital Colorado, University of Colorado, Aurora, Colorado

^q Division of Pediatric Surgery, Hiram C. Polk, Jr, MD Department of Surgery, University of Louisville, Louisville, KY

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ABSTRACT

Objective: Ovarian torsion in pediatric patients is a rare event and is primarily managed by pediatric general surgeons. Torsion can be treated with detorsion of the ovary or oophorectomy. Oophorectomy is the most common procedure performed by pediatric general surgeons for ovarian torsion. The purpose of this systematic review by the American Pediatric Surgical Association Outcomes and Evidence Based Practice Committee was to examine evidence from the medical literature and provide recommendations regarding the optimal treatment of ovarian torsion.

Methods: Using PRISMA guidelines, six questions were addressed by searching Medline, Cochrane, Embase Central and National clearing house databases using relevant search terms. Risks of ovarian detorsion including thromboembolism and malignancy, indications for oophorectomy, benefits of detorsion including recovery of function and subsequent fertility, and recommended surveillance after detorsion were evaluated. Consensus recommendations were derived for each question based on the best available evidence.

Results: Ninety-six studies were included. Risks of ovarian detorsion such as thromboembolism and malignancy were reviewed, demonstrating minimal evidence for unknowingly leaving a malignancy behind in the salvaged ovary and no evidence in the literature of thromboembolic events after detorsion of a torsed ovary. There is no clear evidence supporting the benefit of oophorectomy after a single episode of ovarian torsion. The gross appearance of the ovary does not correlate with long-term ovarian viability or function. Pregnancies have occurred in patients after detorsion of an ovary both spontaneously and with harvested oocytes from previously torsed ovaries. The consensus recommendation for imaging surveillance following ovarian detorsion is an ultrasound at 3 months postprocedure but sooner if there is a concern for malignancy.

Conclusion: There appears to be overwhelming evidence supporting ovarian detorsion rather than oophorectomy for the management of ovarian torsion in pediatric patients. Ovarian salvage is safe and is the preferred treatment for ovarian torsion. Most salvaged ovaries will maintain viability after detorsion.

Type of study: Systematic review of level 3–4 studies.

Level of evidence: 3–4

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* Corresponding author at: Division of Pediatric General and Thoracic Surgery Cincinnati Children's Medical Center, 3333 Burnett Ave., Cincinnati, OH 45229.

E-mail address: Roshni.dasgupta@cchmc.org (R. Dasgupta).

Idiopathic ovarian torsion is a relatively rare event within the pediatric age (<18) group. A vast majority of torsions occur during the reproductive years in women from 20 to 40 years of age [1]. Within the pediatric age group, torsion has an estimated incidence of 4.9:100,000 as noted by an analysis of the 2006 Kids' Inpatient Database (KID) [2]. In their cohort of 1232 patients, the mean age was 14.5 years, and of those with race information available, 51% were Caucasian.

Surgical treatment options for ovarian torsion include detorsion alone, detorsion with oophorectomy, and oophorectomy. Detorsion was initially described in 1946 by Way as a safe and effective treatment for ovarian torsion [3]. Despite this and many other early reports of success with detorsion, a majority of patients continue to undergo oophorectomy. A recent examination of pediatric patients in the National Inpatient Sample (NIS) demonstrated that 15% of patients underwent detorsion, 6% underwent detorsion with oophorectomy, and 78% underwent oophorectomy [4]. In this sample, rates of oophorectomy were higher at nonteaching hospitals, in younger patients, and within the south and rural regions of the United States. The authors also noted that the rate of oophorectomy has not changed despite increasing growing evidence of the benefit and safety of ovarian preservation.

Variation in practice patterns exists between pediatric general surgeons and pediatric gynecologists, with gynecologists more likely to perform ovarian preservation for torsion than pediatric general surgeons [5]. In a single-institutional series of 34 patients, Aziz et al. reported that 6% of ovarian detorsion procedures were performed by a pediatric general surgeon while 94% were performed by a pediatric gynecologist [6]. These observations were also confirmed in a Pediatric Health Inpatient Services (PHIS) study evaluating 43 freestanding children's hospitals, which found that pediatric surgeons were significantly more likely than gynecologists to perform oophorectomy (versus detorsion) [7]. These authors also noted that the rate of oophorectomy has not changed despite increasing evidence for the benefit and safety of ovarian preservation.

This review systematically investigates the existing evidence for managing idiopathic ovarian torsion in the pediatric population. Specifically, the risks and benefits of detorsion are evaluated, as is evidence supporting surveillance practices after operative care. Summary recommendations are presented based on the quality of available evidence. The primary goal of these recommendations is to decrease variability in practice patterns and increase the rate of ovarian preservation.

1. Methods

1.1. Research questions

The American Pediatric Surgical Association (APSA) Outcomes and Evidence Based Practice (OEBP) committee vetted and selected the following questions for this systematic review:

1. What are the risks of ovarian detorsion?
 - a. What is the risk of pulmonary embolism?
 - b. What is the risk of leaving malignancy behind?
2. Is oophorectomy indicated at the time of detorsion?
3. What are the benefits of ovarian detorsion?
 - a. Does recovery of ovarian function correlate with gross appearance at the time of operation?
 - b. Is there evidence of subsequent fertility?
4. What is recommended surveillance imaging after ovarian detorsion?

The initial English language database search was conducted using the Medical Subject Headings (MeSH) terms "ovarian torsion," "ovarian preservation" and "fertility" with all publication dates through February 2015. MEDLINE, Cochrane, Embase, Central, and National Guideline Clearinghouse databases were queried. Inclusive and redundant clinical search terms relevant to each question were then applied to ensure completeness of the literature search. Any additional articles identified

in the references of relevant articles were also included (snowballing methodology). The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines were followed [8].

A PRISMA flow diagram of the search and exclusion process is presented in Fig. 1. The list of 473 titles and/or abstracts generated by the automated search was reviewed independently by two of the authors (RD and ER) and articles were excluded if they did not address any of the study questions. Single case reports, reviews, expert opinions, and animal studies were also excluded.

After these exclusions, 96 studies were allocated to the most appropriate study questions. These were then reviewed for quality of evidence and pertinence to the questions and underwent full review. All articles were Oxford Centre for Evidence-Based Medicine (OCEBM) levels 3 and 4 (Table 1). Selected articles were all retrospective studies with no prospective trials included. There were several meta-analyses, large database studies (PHIS, KID, NIS), and case series noted in the selected literature for review. Each of the 96 articles was assessed for level of evidence, study design, population definition and source, study period, intervention or exposure, comparison measures, outcome measures, sample size, and findings. Critical analysis of each article was performed in accordance with OCEBM guidelines [9].

For each study question, the highest-quality studies were included for analysis; this was done by determining level of evidence and consensus between authors. Articles that addressed more than one question were included. Based upon the best available evidence, a consensus statement of recommendations in response to each study question was then generated.

2. Results

- 1) What are the risks of ovarian detorsion?
 - a. What is the risk of pulmonary embolism?

The risk of pulmonary embolism is often quoted as the primary indication for performing an oophorectomy rather than detorsion. The putative mechanism of thromboembolism is the potential release of thrombi from the vascular pedicle subsequent to ovarian detorsion. McGovern examined the literature and noted two definitive cases of pulmonary embolism (PE) associated with ovarian torsion since 1900; both of those cases were noted to be among 672 patients who underwent oophorectomy. In this report, no cases of PE were noted among the patients who underwent detorsion alone [10]. No case of a thromboembolic event associated with ovarian detorsion has ever been reported in the literature.

2.1. Recommendation

There is no evidence of a risk of pulmonary embolism specifically attributable to ovarian detorsion; therefore, this theoretical risk should not influence operative decision-making. (Level IV evidence; Recommendation Level D).

- b. What is the risk of leaving a malignancy behind?

The incidence of malignancy in ovarian torsion is 2% in adults [11]. The actual incidence of malignancy in the pediatric population is unclear, yet the concern for underlying malignancy is often a driving factor in proceeding with oophorectomy [1,12,13]. The torsed ovary is often enlarged and discolored, and may be difficult to evaluate for the presence of an underlying malignancy [14]. Ten articles were reviewed that examined this question. In these papers, malignancies were detected in 0.4%–5% of resected ovaries. Two case-series described a total of 126 patients with US-confirmed torsion, none of which had any malignant elements found after oophorectomy [15,16]. In a separate case series of 40 patients, 2 (5%) malignant neoplasms were reported (a dysgerminoma and adenocarcinoma), and both were identified during the operation [5]. The 2006 KID database noted 5

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