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Clinical and Radiologic Factors Associated with Adnexal Torsion in Premenarchal and Menarchal Children and Adolescents

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Abstract

Background: Adnexal torsion is a gynecologic emergency in children and adolescents but remains a challenging diagnosis, with no consistent clinical or radiologic diagnostic criteria. Our objective was to identify risk factors associated with adnexal torsion in premenarchal and menarchal patients with surgically confirmed torsion compared with those without torsion.

Methods: We conducted a retrospective chart review of all patients who underwent surgery between January 2016 and December 2019 for possible adnexal torsion. Data on demographics, clinical characteristics, radiologic variables, and operative findings were compared using descriptive statistics. Independent predictors of torsion were then examined in multivariate logistic regression models.

Results: Of the 291 patients who underwent surgery, 168 (57.7%) had torsion. Patients with torsion were younger than those without torsion (11.9 vs. 14.2 years, $P < .01$). Vomiting was significantly associated with torsion for all patients ($P < .001$). Large adnexal volume and absent arterial Doppler flow were associated with torsion for the total population and menarchal subgroup. A logistic regression model for the total population that controlled for age and menarchal status found that vomiting (adjusted odds ratio [aOR] 5.92, 95% confidence interval [CI] 2.87-12.22), highest adnexal volume category (aOR 4.92, 95% CI 2.25-10.75), and absent arterial Doppler flow (aOR 2.674, 95% CI 1.28-5.60) were associated with torsion.

Conclusions: Vomiting, enlarged adnexal volume, and absent arterial Doppler flow were associated with adnexal torsion. However, no single risk factor accurately diagnosed torsion, and multiple factors should be interpreted together.

Keywords: torsion, ovary, adnexa, risk factors, diagnosis

Level of Evidence: Study of Diagnostic Test, Level II

Abbreviations: OR: operating room, BMI: body mass index, CT: computerized tomography, MRI: magnetic resonance imaging

Introduction

Adnexal torsion is a gynecologic emergency in young women and adolescents that involves twisting of the ovarian pedicle and/or fallopian tube that can result in compromised blood flow and viability of the ovary or tube [1]. If not treated promptly, adnexal torsion can potentially have permanent consequences, including loss of the ovary or fallopian tube, diminished hormonal function, and potential impaired fertility. Therefore, prompt diagnosis is imperative.

Despite this, diagnosis prior to surgical evaluation has remained a clinical challenge. The presentation of torsion can vary and also mimics other acute pathology in children and adolescents, such as hemorrhagic ovarian cysts, ruptured ovarian cysts, and appendicitis [2]. Previous studies have examined various clinical, laboratory, and radiologic risk factors, but there are no agreed-upon diagnostic criteria [3-8]. Most of these studies are limited by small numbers, are often purely descriptive, lack a control group of patients without torsion, and rarely differentiate between premenarchal and menarchal patients [9].

The consequences of being unable to accurately predict adnexal torsion are risk of prolonged ischemia, which can affect long-term ovarian and tubal function, as well as exposure of patients to unnecessary surgery. Given that the rate of intraoperative diagnosis of torsion is as low as 30% but a missed diagnosis can have significant clinical implications [10], improved diagnostic criteria are needed. This would more accurately identify which patients need surgery, improve rates of ovarian salvage and long-term reproductive function, and prevent unnecessary surgery. The objective of this study is to identify and test previously identified clinical and radiologic risk factors associated with adnexal torsion in both premenarchal and menarchal children and adolescents in a large population of patients with surgically confirmed torsion compared with those with surgically confirmed absence of torsion.

Methods

Data Collection

A multidisciplinary team including Pediatric Gynecology, Pediatric Surgery, and Pediatric Radiology conceptualized and designed this study. We conducted a retrospective chart review of all patients who went to the operating room (OR) between January 1, 2016, and December 31, 2019, at Nemours Children's Health because of concern for ovarian or adnexal torsion. Surgeries were performed at Nemours Children's Hospitals in Wilmington, Delaware, and Orlando, Florida, and two affiliated community hospitals. Eligible patients were identified from hospital billing and coding databases using procedure codes and confirmed by chart review. Inclusion criteria were patients who were assigned female sex at birth, had abdominal or pelvic imaging, and went to the OR for suspicion of torsion. Data were abstracted from the electronic medical record by chart review and entered into Research Electronic Data Capture (REDCap) forms [11,12]. Abstracted data included patient demographics (age, race, ethnicity, insurance), clinical characteristics (weight, body mass index [BMI], pubertal and menarchal status), clinical symptoms, radiologic reports and imaging, and operative reports. Pubertal status was defined as whether or not patients had started any pubertal development, such as breast or axillary hair development, and was based on any explicit statement about pubertal changes or timing or documented Tanner staging. Menarchal status was defined as whether patients had undergone menarche and based on documentation of menses or a last menstrual period. Images from reports with missing data were obtained and reviewed by two radiologists.

The dependent variable was the presence or absence of adnexal torsion at the time of surgery, as defined by reviewing operative reports. Potential independent variables for the prediction of

torsion included demographic, clinical, and radiologic risk factors that were identified based on prior studies and the investigators' clinical experience. Demographic characteristics included age, race, ethnicity, BMI, pubertal status, and menarchal status. Included clinical factors were history of an ovarian or adnexal cyst; history of surgery for an ovarian or adnexal cyst; history of prior adnexal torsion; time since last menstrual period for menarchal patients; and presence, nature, duration, and severity of pain, nausea, and vomiting.

Radiologic variables were presence, location, and volume of an adnexal cyst or mass; adnexal ratio; peripheralization of ovarian follicles; presence of pelvic free fluid; presence of arterial and venous Doppler flow in the affected adnexa; and the radiologist's impression related to torsion. The location and source of the adnexal cyst as ovarian, paratubal, or adnexal/pelvic was based on the radiologic report. If this was not explicitly stated in the report or there was uncertainty, the images were reviewed by our radiologists to make this determination. Adnexal volume was defined as the volume of the entire ovary or of an adnexal or pelvic cyst if it was separate from the ovary. In addition to reporting the absolute volume, the volumes were also stratified into previously defined statistically derived categories [9]. These categories are < 105 ml and ≥ 105 ml for menarchal patients and < 6 ml, 6-17 ml, and > 17 ml for premenarchal patients. Adnexal ratio was defined as the ratio of the affected adnexa to the unaffected adnexa. This was not able to be calculated if there was an adnexal or pelvic cyst separate from the ovary or if the opposite ovary was not present or unable to be visualized or measured. The ratio was also divided into previously defined categories [9]: < 1.25, 1.25-21, and > 21 for premenarchal subjects and < 2, 2-21, and > 21 for menarchal subjects.

The radiologic impression related to torsion was also investigated as a potential independent variable, as it is the factor that is typically relied on at our institution to make a diagnosis of

torsion and often drives management, although it is subjective. The radiologic impression was categorized based on the written impression of the ultrasound by the original reading radiologist. This was not changed by the radiologists involved with this study. The impression was defined as torsion when the report mentioned that the ultrasound findings were consistent with or concerning for torsion or that torsion could not be ruled out. It was classified as no torsion only if the report specifically stated that torsion was not present. Ultrasound reports in which there was no mention of an interpretation related to torsion were classified as "no impression related to torsion." Potential radiographic factors specific to computerized tomography (CT) and magnetic resonance imaging (MRI) studies were also abstracted. With these studies, differences in adnexal size, signal, enhancement, and diffusion were based on whether these factors were mentioned in the study report.

Data Analysis

Frequency counts, percentages, means, and standard deviations were used to describe the study sample. Variables were compared between patients with and without surgically diagnosed torsion for the entire study population, as well as within premenarchal and menarchal subgroups. Continuous variables were compared using two-sample *t*-tests or Mann-Whitney U tests for non-normally distributed variables. Chi-square or Fisher exact tests were used to compare categorical variables, as appropriate. Missing data were excluded from analysis of individual variables.

Independent predictors of torsion were then examined in multivariate logistic regression models for the overall population and separately for the premenarchal and menarchal subgroups. A decision was made not to include the radiologic impression in the logistic regression models because of its subjectivity and lack of reproducibility at other institutions. These models were then adjusted for possible confounding variables of age, race, and BMI. In an effort to refine the

estimates and create a single model for the overall population, we also examined logistic regression models for the total population that controlled for age and menarchal status, as well as interaction terms for those variables. All variables were assessed for multicollinearity. A p-value of < 0.05 was considered statistically significant. Statistical analyses were performed using IBM SPSS Statistics for Windows, version 26 (IBM Corp., Armonk, NY, USA). This study was approved by the Nemours Children's Health System Institutional Review Board.

Results

During the study period, 291 patients underwent surgery because of concern for adnexal torsion. Of these 291 patients, 168 (57.7%) were found to have torsion. Demographic and baseline clinical characteristics for all patients and by torsion vs. non-torsion subgroups are presented in Table 1. The mean age of all patients was 12.8 (SD 3.6) years, with a range of 0 to 18 years. Patients who had surgically confirmed torsion were younger than those without torsion (11.9 vs. 14.2 years, $P < .01$). There were no differences in race or ethnicity between the groups. The majority of patients fell within the normal weight and BMI ranges, although approximately one-third of patients were overweight or obese, with no differences between groups. While the majority of surgeries occurred in menarchal (71.8%) females and especially in those who had started pubertal development (82.8%), prepubertal and premenarchal patients who underwent surgery were significantly more likely to have adnexal torsion than not to have torsion ($P < .001$): 87.5% of the prepubertal and 83.3% of the premenarchal patients who went to the OR were confirmed to have torsion vs. only 51.4% of the pubertal and 47.8% of the menarchal and pubertal cohorts.

Potential clinical risk factors for adnexal torsion are presented in Table 2. In terms of clinical history and presentation, patients who had a history of an ovarian or adnexal cyst and those with a history of prior adnexal surgery were less likely to have torsion ($P = .029$ and $P = .021$, respectively). Patients who had experienced a torsion in the past were not more or less likely to have a repeat torsion. Adnexal torsion was not associated with the presence, nature, duration, or severity of pain. The presence of both nausea ($P < .001$) and vomiting ($P < .001$) was associated with torsion. While both nausea and vomiting were associated with torsion in menarchal patients ($P = .002$ and $P < .001$, respectively), only vomiting was significantly associated with torsion in premenarchal patients ($P = .004$).

Radiologic factors were then examined (Table 3). Almost all patients (97.9%) had an ultrasound performed as part of their evaluation. A quarter of patients had a CT scan and 13.4% had an MRI study. More than half of patients (60.9%) had an adnexal cyst or mass visualized on imaging. Of those with a cyst or mass, the majority were characterized as ovarian on imaging. There were a small number of paratubal cysts, and some cysts identified only as adnexal or pelvic in origin. The presence and anatomic location of a cyst were not associated with torsion. Peripheralization of ovarian follicles or the presence of pelvic free fluid was also not associated with torsion. Absence of both ovarian arterial ($P < .001$) and venous ($P = .003$) Doppler flow, as well as the radiologic impression related to a diagnosis of torsion on ultrasound ($P = .004$), was found to be significantly associated with torsion. No CT or MRI findings were associated with torsion.

Radiologic data were then broken down into premenarchal and menarchal subgroups because of the natural age-related differences in normal ovarian size and presence of physiologic cysts (Table 4). The presence or location of adnexal cysts or masses, peripheralization of ovarian follicles, and presence of pelvic free fluid were not associated with torsion in either subgroup.

The median adnexal volume was significantly larger in the menarchal group (P < .001) but not the premenarchal group with torsion (P = .42). When volume was stratified into previously derived categories for each subgroup, it was associated with torsion in both groups (P = .012 for premenarchal patients and P < .001 for menarchal patients). The median adnexal ratio of volumes of the affected to unaffected adnexa was similarly significant only for menarchal patients (P = .008). Use of the previously derived adnexal ratio categories yielded the same results, with a significant association only for menarchal patients (P = .009). Absence of ovarian arterial (P < .001) and venous (P = .039) Doppler flow was found to be associated with torsion only in the menarchal group. Similarly, the impression of the radiologist in regard to torsion on ultrasound was associated with torsion only in the menarchal group (P = .016).

Unadjusted multivariate logistic regression analyses showed that vomiting, having an adnexal volume in the largest category, and absent arterial Doppler flow were associated with torsion in the overall population and the menarchal subgroup (Table 5). In the premenarchal subgroup, only vomiting was associated with torsion. When adjusted for potential confounding variables of age, race, and BMI, the same variables remained significantly associated with torsion in the overall population and the menarchal subgroup, with the addition of age for the total population. In addition, adnexal volume was found to be more significantly associated with torsion for menarchal patients. In the premenarchal group, no variables retained significance. For the overall population, a single model controlling for age and menarchal status retained the same pattern of significance, with vomiting (aOR 5.92, 95% CI 2.87-12.22, P < .001), highest adnexal volume category (aOR 4.92, 95% CI 2.25-10.75, P < .001), and absent arterial Doppler flow (aOR 2.674, 95% CI 1.28-5.60, P < .01) all significantly associated with adnexal torsion. Testing for

multicollinearity resulted in variance inflation factors of less than 2, indicating the absence of multicollinearity that could affect the models.

Discussion

Although there are many descriptive studies on adnexal torsion in children and adolescents, there is ongoing controversy about how best to make this challenging diagnosis. Previous studies were often small, lacked a control group, and rarely treated premenarchal and menarchal patients as potentially different populations. In addition, many studies did not incorporate both clinical and radiologic variables.

Our study examined both clinical and radiologic risk factors in a large population of children and adolescents with and without adnexal torsion. Premenarchal and menarchal subgroups were also analyzed separately because of inherent differences in the size of reproductive structures in these different age groups. For the total population, nausea, vomiting, both absent arterial and venous Doppler flow, and the radiologic impression related to torsion were all significantly associated with torsion. However, when these factors were reexamined for the premenarchal and menarchal subgroups separately, only vomiting was associated with torsion in premenarchal patients. This association was no longer seen for premenarchal patients in adjusted logistic regression models, but vomiting remained significantly associated with torsion for the total population, menarchal subgroup, and in the single adjusted model that incorporated menarchal status to fit the entire population. While most previous studies did not separate nausea and vomiting as risk factors, a prior study that did found similar findings with only vomiting as a significant risk factor for torsion [9].

In clinical practice, radiologic findings suggestive of possible torsion can be deceiving and inconsistent but are often relied upon to evaluate abdominal pain in adolescents [6-8]. A recent study in patients with a wide range of ages demonstrated that common ultrasound findings seen in cases of proven torsion included an enlarged ovary, whirlpool sign, ovarian stromal edema, and free fluid in the pelvis [13]. While previously thought to be necessary in the evaluation of torsion, the absence of Doppler signals was not a common sign. Other studies have confirmed the limited clinical utility of Doppler flow in the evaluation of adnexal torsion [4,7,8,13,14], but this continues to be used clinically. A recent study proposed that an algorithm including multiple sonographic features such as intra-ovarian flow, peripheral follicles, volume of both ovaries, and presence of cysts or masses performs better than relying on single features alone [15]. Another recent study of adolescents with surgically confirmed adnexal torsion found that 54% of patients had a cyst or mass in the affected adnexa [16]. This is similar to our findings. However, almost all their patients were menarchal. When our patients were divided into subgroups based on menarchal status, the presence of cyst or mass was more likely in the menarchal group (67%) than in the premenarchal group (42%), although this finding was not associated with torsion in either subgroup. This study also found that peripheralization of follicles and presence of free fluid were associated with torsion in those patients who did not have an adnexal mass. Our study did not find that the presence or location of an adnexal cyst or mass, peripheralization of ovarian follicles, or presence of free fluid was associated with torsion.

In terms of size-specific radiologic findings, adnexal volume and ratio were associated with torsion in menarchal patients, but only a volume of more than 17 ml was associated with torsion in the premenarchal group. This was confirmed in adjusted logistic regression models, and the association with adnexal ratio disappeared for the menarchal group in adjusted regression

models. While an ovarian dimension of 5 cm is often quoted as a risk factor for torsion [10,17], a previous study in adolescents suggested a volume threshold of more than 75 ml as predictive of torsion [7]. No studies have reported volume thresholds in premenarchal patients. We used previous statistically derived volume and adnexal categories for our study, which may have underestimated findings given their larger values than other previously published data.

The presence of Doppler flow has not been found to be associated with torsion in many other studies. Our findings on Doppler flow differed somewhat from other studies in finding that absent arterial and venous Doppler flow was associated with torsion for the total population and in the menarchal subgroup. Absent arterial Doppler flow remained significantly associated with torsion in the total population and menarchal subgroup in the logistic regression models. This difference in absent arterial Doppler flow as a risk factor in menarchal vs. premenarchal patients is interesting and is most likely related to the dual blood supply to the ovary, especially in larger, well-developed ovaries, and the normal finding of low diastolic flow in small, prepubertal ovaries [18], but it deserves further study. However, while Doppler flow was more likely to be absent in the torsion group and present in the non-torsion group, arterial Doppler flow was only accurate in 61% of patients, including only 65% of menarchal patients. Therefore, the presence or absence of Doppler flow should be interpreted cautiously and should not guide decision-making alone.

Similarly, while the ultrasound interpretation by Radiology was associated with torsion in both the overall population and especially in menarchal patients, this impression was correct for only 54% of the total population and 51% of the menarchal patients, with 21% of all torsions explicitly missed and 48% of patients without torsion incorrectly diagnosed as having torsion. This is similar to a prior finding that ultrasound had 51% sensitivity for a diagnosis of torsion

[10]. We suspect that the association of the radiologic interpretation in menarchal patients is related to the discrepancy in findings on Doppler flow between the menarchal and premenarchal subgroups. However, as the radiologic interpretation is subjective, it is difficult to draw conclusions from this information.

Some radiologists have recently advocated for the use of MRI to help make a diagnosis of torsion, either as a first-line imaging modality or as an adjunct to equivocal ultrasound findings [19]. We did not find any MRI or CT findings that were associated with torsion. However, while CTs were likely all done with a similar protocol, the MRIs were not and this would affect the potential ability to diagnose torsion. Although the cost of MRI is high, it is increasingly being used in pediatric emergency rooms to evaluate for possible appendicitis in order to avoid radiation exposure in pediatric patients [20,21]. Evaluation of the ovaries and adnexa can be obtained at the same time, with the appropriate protocol. Further research is needed to evaluate the clinical utility of MRI for the diagnosis of adnexal torsion, especially in situations in which the pre-test probability of torsion is intermediate, while balancing the increased costs and extra time. In summary, we would argue that while radiologic findings are helpful in making a diagnosis of possible torsion, this remains a clinical diagnosis.

A previous study used prospective data to create a composite score to predict adnexal torsion in both premenarchal and menarchal children and adolescents. They found that vomiting, adnexal volume, and adnexal volume ratio were independent predictors of torsion in both groups of patients. Duration of pain, intermittent pain, nausea, and absence of arterial or venous flow were not associated with torsion [9]. However, this study was limited by the small number of patients with confirmed torsion. A follow-up study at a different institution with a more heterogenous patient population retrospectively applied this composite score to 59 patients with possible

adnexal torsion, 53 of whom were confirmed to have torsion intraoperatively. This study confirmed many of the findings of the previous study, including vomiting, adnexal volume, and adnexal ratio, although it also found presence of an adnexal mass and ovarian edema to be associated with torsion. Presence of arterial and venous Doppler flow was not associated with torsion [22]. Our study confirms the previous study's finding of vomiting as the most important clinical predictor of torsion. We used the same adnexal volume and ratio categories but found only volume to be associated with torsion.

Strengths of this study are its large patient population and the presence of a robust control group. This study also involved Pediatric Gynecology, Surgery, and Radiology from its inception to obtain all relevant viewpoints and investigate a wide variety of potential risk factors. Radiology was able to review the actual images to more accurately obtain data on variables of interest. The major limitation was missing data because of the retrospective nature of the study. Although all efforts were made to obtain missing clinical information, especially if the patient was initially seen at an outside hospital, our data relied on adequate clinical investigation and documentation. Similarly, while almost all missing radiographic images were able to be obtained and reviewed by our radiologists, the imaging techniques were variable across institutions, and some of the imaging was inadequate and was therefore not able to answer our research questions. Another limitation of this study is that the study population was limited to those who underwent surgery for suspicion of possible adnexal torsion. This decision may have caused exclusion of patients who were evaluated as an outpatient or in the emergency department and discharged home without surgical exploration. Conversely, inclusion of all patients evaluated for abdominal pain would similarly miss those who were discharged without surgery or even imaging. We chose our approach to allow a comparable control group of those needing surgical management due to

suspicion for torsion but who were subsequently found not to have torsion because we think they present the most relevant diagnostic challenge. However, the proper interpretation of this study is risk factors for torsion in patients who were determined to require surgical intervention and not necessarily all those who present for evaluation for abdominal pain. Lastly, while this study included patients from four different hospitals in three different states and surgery was performed by two different surgeon groups, it was done under a single health system, which may limit its generalizability.

Conclusion

This study is an addition to the growing body of literature on the diagnosis of adnexal torsion in adolescents. It confirms the importance of vomiting and enlarged adnexal volume as the most important predictors of torsion. It also raises the question of whether absent arterial Doppler flow may be useful in helping to make the diagnosis in menarchal patients only. However, no one factor alone has been found to accurately diagnose adnexal torsion, and these finding should be used in combination. Further evaluation of our data and future prospective studies with multi-institutional collaboration are needed to validate or refine a previously created composite score that incorporates multiple risk factors. This will allow for practical use of such a diagnostic tool in clinical settings to help triage children and adolescents with abdominal or pelvic pain to appropriate clinical or surgical care.

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