

Comparison of underlying lesions in pediatric and adult ovarian torsion

Safiye Aktas, MD, Seyran Yigit, MD, Gulden Diniz, MD, Fatma S. Pehlivan, MD, Ragip Ortac, MD.

ABSTRACT

Objective: To compare the pediatric and adult ovarian torsion and explore a quantitative value to predict a possible underlying tumor.

Methods: This study included 32 pediatric and 33 adult female cases diagnosed with ovarian torsion and underwent surgical treatment in Dr. Behçet Uz Children's Research Hospital and Atatürk Education and Research Hospital, Izmir, Turkey between 1989 and 2005. We evaluated the properties of the cases statistically.

Results: The mean age of pediatric was 8 years and 3 months and adult cases was 39 years and 8 months. Six cases had an underlying tumor in both pediatric and adult group. The mean diameter and volume difference were 6.84 cm, 91 cc in pediatric and 12.69 cm and 1087 cc in the adult group. In statistical analysis, the diameter and volume increase were significantly higher in cases with underlying

tumor in pediatric group. The cut-off value was 7 cm in diameter and 104 cc in volume increase. In the adult group, the diameter and volume increase were not significant in tumor positive and negative group.

Conclusion: Torsion of the ovary requiring surgery, is rare and is the most common reason of abdominal/pelvic mass. These cases are often difficult to decide for surgical procedure especially in pediatric cases. We conclude that an underlying lesion more commonly occurs in children with an increase in ovarian volume of more than 104 cc and a diameter more than 7 cm. Great care should be taken for laparoscopic conservative management especially in these cases. The quantitative analysis is not predictive for the underlying solid lesion in adult cases.

Saudi Med J 2006; Vol. 27 (8): 1183-1186

Ovarian torsion, which causes formation of mass, is very rare and is the most common reason of abdominal/pelvic mass seen in premenarchal girls undergoing ovarian surgery. Contrary in adult patients the main cause of the ovarian torsion is prior pelvic surgery.¹ Alternatively, ovarian torsion is not very rare in fetus and perinatal ovarian torsion is the most common complication of the fetal ovarian cysts.^{2,3} Torsion of the normal ovary results from the excessive mobility of the ovary. Associated ovarian pathologies, such as cyst of tumors may lead to excessive rotation causing torsion.⁴ The salvage rate

in cases of torsion is due to delays in diagnosis and intervention.⁵ The aim of this study was to review and compare the pediatric and adult ovarian torsion cases and determine its relationship with benign or malignant, cystic or solid tumors and explore a quantitative value to predict a possible underlying tumor in a fully necrotic material.

Methods. This study included 65 females who underwent unilateral salpingo-oophorectomy procedures due to ovarian torsion in 2 research hospitals between 1989 and 2005. The hematoxylin-

From the Department of Pathology (Aktas, Diniz, Ortac), Dr Behçet Uz Children's Research Hospital and the Department of Pathology (Yigit, Pehlivan), Atatürk Education and Research Hospital, Izmir, Turkey.

Received 25th February 2006. Accepted for publication in final form 26th April 2006.

Address correspondence and reprint request to: Dr. Safiye Aktas, 1420 sok. No 67/1 35230, Alsancak, Izmir, Turkey. Tel. +90 (232) 4641045. Fax. +90 (232) 3625522. E-mail: safiyeaktas@yahoo.com

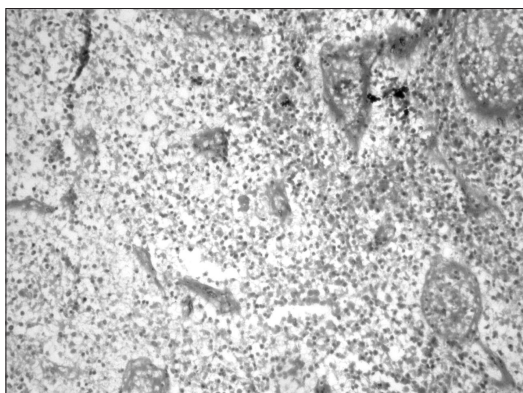


Figure 1 - Torsed ovary section with endodermal sinus tumor (Hematoxylin and Eosin x 100).

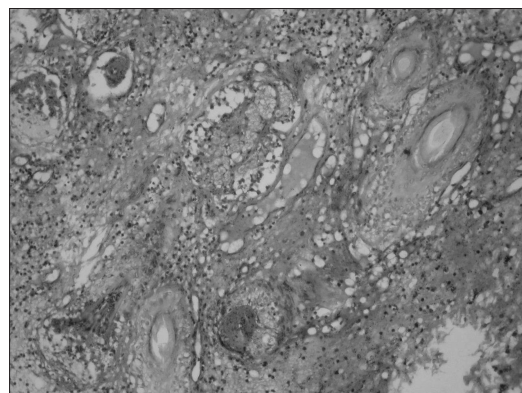


Figure 3 - Well preserved epidermal appendices in a torsed ovary with teratoma (Hematoxylin and Eosin x 100).

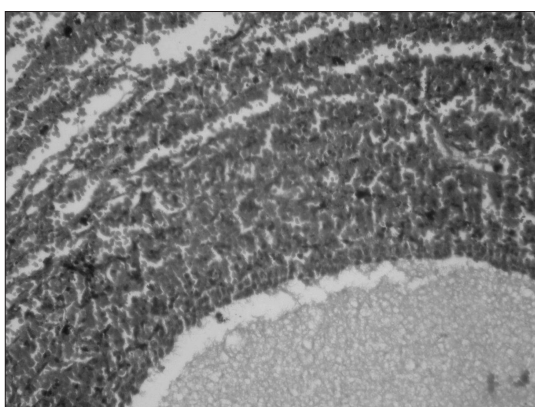


Figure 2 - Benign epithelial cyst in torsed ovary (Hematoxylin and Eosin x 100).

eosin stained slides (mean 4 [1- 10] slides) of the cases with ovarian torsion were retrospectively reviewed by 2 pathologists. Age of the patient, side of the lesion, underlying tumor, localization, macroscopic properties and diameter were recorded for each patient. Simple or follicular cysts, torsion with cyst formation were considered as having no underlying tumor. Volume was calculated by: $V=1/2$ length x width x depth (a simplified formula for a prolate ellipse).⁵ Increase in volume was assessed by the difference of the volume of the ovary and the mean expected value for the age of each case.⁵ The parameters were evaluated by Chi square, t test and Mann Whitney U test. Statistical analysis was completed by using SPSS version 3.0. The p value of <0.05 were considered statistically significant.

Results. The mean age of the pediatric patients was 8 years and 3 months (range: 1 month - 16 years) and adult cases was 39 years and 8 months (range: 18-75 years). Three cases had underlying solid tumors diagnosed dysgerminoma, endodermal

sinus tumor (**Figure 1**) and mixed germ cell tumor, while in 14 cases cysts (**Figure 2**) were detected and in 3 cases mature cystic teratoma (**Figure 3**) were diagnosed. In the other 12 cases, no underlying lesion were detected in pediatric group. In adult group a 70 years old woman was diagnosed as granulosa cell tumor, 4 cases had serious benign tumors; a 37 years old woman had mature cystic teratoma. In 23 cases, torsed benign ovarian cysts were diagnosed and in 4 cases no underlying lesion were detected. In a 3-year-old case, embryonal carcinoma and endodermal sinus tumor areas were available to detect in some sections. The lining epithelium of benign cysts was detectable in some cases. The main preserved tissue in mature cystic teratoma was hair follicles of epidermis with no necrosis. Cystic areas including keratin were also observed. Some of the cases included multiple cysts, which were suspected as polycystic ovary. In 3 pediatric cases, ovarian torsion occurred due to an intraabdominal pathology such as strangulated inguinal hernia. The hemorrhagic necrosis was demonstrated in large fields. Eighteen cases were right sided and 14 were left in pediatric group, while 14 were right sided and 15 were left in adult group. In 4 adult cases, the side were not recorded. The mean macroscopic diameter and volume difference of the torsed ovary were 6.84 ± 2.75 cm and 91 ± 85.3 cc in the pediatric group and 12.69 ± 4.89 cm and 1087 ± 1407 cc in the adult group. All cases included hemorrhagic necrosis except one early operated case with massive edema. Hemorrhagic necrosis was also seen in tuba uterine of the cases. In statistical analysis, the ratio of the underlying lesion did not differ between pediatric and adult cases ($p=0.626$, Chi square), although the mean diameter, which was proportional to the age of the groups, was higher in adult cases ($p=0.003$, t-test). In the pediatric group, the diameter and volume increase were significantly

higher in cases with the underlying tumor (solid tumor and mature cystic teratoma) positive cases than negative cases (Mann Whitney U test, $p=0.02$ and $p=0.01$). The mean diameter in the tumor positive group was 9 cm and 6 cm in the tumor negative group. The calculated cut-off values were 7 cm for diameter and 104 cc for volume increase. In the adult group, the diameter and volume increase were not significantly different in tumor positive and negative group ($p=0.161$ and $p=0.261$). The side of the ovary was not related with the underlying tumor in either pediatric or adult group (Chi square, $p=0.365$ and $p=0.682$).

Discussion. Clinicians search for pre-operative markers for the diagnosis of ovarian torsion. However, the pathologists not only search for the diagnosis of ovarian torsion, but also need markers to guess about an underlying lesion or tumor in a fully necrotic torsed ovarian tissue.⁶ There are many studies carried out to investigate a possible role of several things as markers for the diagnosis of ovarian torsion. For example, Cohen et al⁶ found that elevated serum interleukin 6 concentrations were significantly associated with the occurrence of ovarian torsion. Similarly increased incidence of adnexal torsion was found as a complication after gonadotrophin ovulation induction for in vitro fertilization and ovarian hyper stimulation syndrome. Hormonal induction generally causes the ovary to have multiple larger follicle cysts and the presence of huge cysts increases the incidence of ovarian torsion.^{6,7} Most studies examining adnexal torsion suggest that benign ovarian neoplasms are more likely to undergo torsion than malignant ovarian neoplasm. Benign lesions have a 13-fold increased risk of occurring adnexal torsion when compared with malignant tumors. The histological type of the neoplasm does not appear to affect the rate of torsion. The benign tumors with torsion include ovarian cysts, teratomas and rarely cavernous hemangiomas.^{8,9} In a series reported by Brown et al,¹⁰ childhood ovarian masses in order of frequency were as follows: teratomas, simple or epithelial cysts, torsion with cyst formation, germ cell tumors, epithelial tumors, granulosa cell tumors. Leukemia infiltration, metastatic Wilms tumor, uterine neuroblastoma and fibroma might be observed as well. In another series of 140 pediatric cases with ovarian mass, Thempleman et al¹¹ found that patients with ovarian cysts and mature cystic teratomas were statistically more likely to have torsion than those with other benign or malignant tumors. If simple cysts are larger than 5 cm in diameter, the risk of torsion

may be significant. However, the ischemic tissue can act as a trigger for a consumptive coagulopathy that could have a lethal outcome even the torsion occurs from a benign lesion.^{1,10-12} In the series reported by Özcan et al,¹³ 91% of the cases with ovarian torsion had additional adrenal pathology. In 80% of the cases, hemorrhagic necrosis were found and in 20% of them adnexal preservation was possible. A torsed ovary that does not include any cyst or solid tumor may become a solid mass due to congestions, fibrosis, and hemorrhage. Secondary cyst formation might be observed as well. The histological view of a torsed ovary is hemorrhagic infarction. Differential diagnosis of the pediatric ovarian tumors is sometimes difficult even if they are not necrotic. For example, pure endodermal sinus tumor is generally confused histologically with embryonal carcinoma. The tumor in a torsed ovary shows large areas of necrosis with little chance of defining live tumor cells. They are generally in ghost appearance, and the differential diagnosis becomes more difficult.^{1,3,10,14} In a series of 87 women with a mean age of 32 years; 25% had a history of an ovarian cyst and 40% had prior pelvic surgery.¹⁵ In that report, 89% of the cases had enlarged ovary (>5 cm in diameter) while in our cases 95%. In our pediatric age group, 13.6% of the cases had a solid tumor and 63.6% of them had a cystic background. In our series, there are no side predominance and the side of the ovary is not related with the underlying tumor in neither pediatric nor adult group, although it is reported that the right side was affected more frequently than the left.¹⁶ The age distributions of our 32 pediatric ovarian torsion cases are interesting also indicate a possible hormonal stimulus. These cases are whether under 2 years of age or over 8 years. This shows a relationship between hormonal stimuli, maternal and pubertal or prepubertal. Defining an underlying lesion in a torsed ovary is very difficult as discussed. In this study, we compared the properties of pediatric and adult ovarian torsions and explored a quantitative value for diameter and increase in volume to predict a possible underlying tumor. We conclude that an underlying lesion more commonly occurs in children with an ovarian volume increase more than 104 cc and diameter more than 7 cm. Great care should be taken for laparoscopic conservative management especially in these cases although it is reported that adnexal-sparing surgery should be encouraged if possible.¹⁷ Evaluation of ovarian torsion is still a problem in clinical evaluation in pediatric cases.¹⁸ The quantitative analysis is not predictive for underlying solid lesion in adult cases.

References

1. Quint EH, Smith YR. Ovarian Surgery in premenarchal girls. *J Pediatr Adolesc Gynecol* 1999; 12: 27-29.
2. Bagolan P, Giorlandine C, Nahom A, Blancioni E, Truccii A, et al. The management of fetal ovarian cysts. *J Pediatr Surg* 2002; 17: 25- 30.
3. Katz VL, McCoy MC, Kuller JA, Hansen WF, Watson WJ. Fetal ovarian torsion appearing as a solid abdominal mass. *J Perinatol* 1996; 16: 302-304.
4. Garel L, Dubois J, Grignon A, Filiatraut D, Vliet GV. US of the pediatric female pelvis: A clinical perspective. *Radiographics* 2001; 21: 1393-1407.
5. Celik A, Ergun O, Aldemir H, Ozcan C, Ozok G, Erdener A, et al. Long-term results of conservative management of adnexal torsion in children. *J Pediatr Surg* 2005; 40: 704-708.
6. Cohen SH, Wattiez A, Stackheim D, Seidman DS, Lidor AL, et al. The accuracy of the serum interleukin 6 and tumor necrosis factor as markers of ovarian torsion. *Hum Reprod* 2001; 16: 2195-2197.
7. Gorkemli H, Camus M, Clasen K. Adnexal torsion after gonadotrophin ovulation induction for IVF OR ICSI and its conservative treatment. *Arch Gynecol Obstet* 2002; 267: 4-6.
8. Summerville M, Grimes DA, Koonings PP, Campbell K. Ovarian neoplasm and the risk of adnexal torsion. *Am J Obstet Gynecol* 1991; 164: 577-578.
9. Mirilas P, Georgiou G, Zevgolis G. Ovarian cavernous hemangioma in an 8-year-old girl. *Eur J Pediatr Surg* 1999; 9: 116-118.
10. Brown MF, Hebra A, McGeehin K, Ross AJ. Ovarian masses in children: a review of 91 cases of malignant and benign masses. *J Pediatr Surg* 1993; 28: 930-933.
11. Templeman C, Fallat ME, Blinchevaky A, Hertweck SP. Noninflammatory ovarian masses in girls and young women. *Obstet Gynecol* 2000; 96: 229-233.
12. Karl S, Sen S, Zechariah N, Chacko J, Thomas G. Torted ovarian cyst with lethal bleeding diathesis in an infant. *Pediatr Surg Int* 1999; 15: 145-146.
13. 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.
14. Gribbon M, Ein SH, Mancor K. Pediatric malignant ovarian tumors: a 43-year review. *J Pediatr Surg* 1992; 27: 480-484.
15. Henry D, Abbott JT. Ovarian torsion: a fifteen-year review. *Ann Emerg Med* 2001; 18: 156-159.
16. Carvalho JP, Diegoli MS, Carvalho FM, Diegoli CA. Adnexal torsion following gonadotropin-releasing hormone analog therapy: a case report. *Rev Hosp Clin Fac Med Sao Paulo* 2004; 59: 128-130.
17. Powell JK. Benign adnexal masses in the adolescent. *Adolesc Med Clin* 2004; 15: 535-547.
18. Breech LL, Hillard PJ. Adnexal torsion in pediatric and adolescent girls. *Curr Opin Obstet Gynecol* 2005; 17: 483-489.