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Ovarian torsion in pediatric patients: a single-institution case series and mini-review of the literature

Abstract

Ovarian torsion is an infrequent cause of acute abdominal pain in children. In these patients its etiology, diagnosis and treatment may differ significantly from that of adults. We show here our experience in 28 patients aged 9-16 during a seven years time frame (2008-2014). All patients underwent trans-abdominal Ultrasonography (US) associated to Color Doppler Ultrasonography (CDU) and, in some selected cases, second level imaging methods. The surgical approach included open surgery or laparoscopy, either conservative (detorsion plus cystectomy/tumorectomy plus oophoropexy whenever appropriate) or radical, according to patient's conditions. Clinical presentation is non-specific and this frequently leads to diagnostic delay that might cause adnexal infarction. In case of acute abdominal pain, US is the most useful diagnostic tool, coupled with color-Doppler for the evaluation of blood circulation in the affected ovary.

Keywords: Oophoropexy, Ultrasonography, Acute abdominal pain, Adnexal torsion

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Abbreviations: AAP, Acute Abdominal Pain; US, Trans-Abdominal Ultrasonography; CDU, Color Doppler Ultrasonography; CT, Computed Tomography; MRI, Magnetic Resonance Imaging; VLP, Video Laparoscopy

Introduction

Ovarian torsion, also known as adnexal torsion, is an infrequent cause of acute abdominal pain (AAP) in adult women (2.7% of cases of acute gynecologic complaints) which typically affects the ovary and the fallopian tube.[1] In most cases it is caused by a reduced venous return from the ovary, usually consequent to stromal edema, internal hemorrhage or presence of an obstructing mass.[1] However, ovarian torsions represent a rare cause of AAP in children, since the etiology, presentation, diagnosis and management often differ from those in adults.[2,3]

Nearly three-fourths of all cases of ovarian torsion occur during the reproductive years from 20 to 40 years old.[4] In particular, it can occur at any age, but it is more frequent in the early fertile years. The proportion of patients younger than 30 years is approximately 70-75%.[4] Two groups of women show a significantly higher predisposition to ovarian torsion: women in their mid 20s and postmenopausal women. Only 15% of all torsions occur in pediatric patients,[5] with two peaks of incidence, in newborns and at menarche.[6] According to some authors, the latter girls are at risk maybe because of the changes in the weight of their maturing adnexa.[7] Ovarian torsion is extremely rare between the neonatal period and early childhood.[8-12] The real incidence of ovarian torsion in children is currently unknown; in 1996 fewer than 300 cases were present in the world's literature.[13] Moreover, large series from pediatric centers report a case-experience of between 0.3 and 3.5 patients per year.[2]. More recent publications provide an estimation of the incidence of this condition of 4.9 per 100,000 among females 1-20 years old[14] with a mean age showing ample variation according to different studies, from 9 to 14 years old.[3,5,15-17] Despite the difficulty to recognize this condition in children – due to their low frequency – these girls show a more favorable prognosis: the ovarian salvage rate has been reported as low as 10% in adults but raises to 27% in a study regarding pediatric patients.[9] In this perspective, the early diagnosis of ovarian torsion is paramount and will reduce the risk of complications and increase the probability of ovarian preservation. Indeed, some authors report an 80%-90% rate of ovarian salvage with early surgical intervention.[18] Delay and misdiagnosis of adnexal torsion is common in pediatric patients and may cause the loss of the ovary, of the fallopian tube, or both.[19] This is not only attributed to the loss of blood supply during torsion, but also to the oxidative stress during ischemia/reperfusion injury.[20]

The twisting of the ovary may involve a normal ovary (15-25%)[21,22] or an ovary with functional pathology, benign or malignant neoplasm;[6,13] however, malignancies are very rare. At presentation, the symptoms are usually nonspecific, thus often causing a delay of some hours in both diagnosis and surgical management.[22] A timely diagnosis is indeed necessary to prevent infarction of the adnexal structures. Moreover, to date there is not a reliable and univocal method to assess a correct diagnosis, thus it is not infrequent to have a definitive diagnosis during surgery.[23,24]

In the present report we retrospectively review 29 cases of ovarian torsion treated in our pediatric surgical department over a 7-years

period. The purpose of this study is to illustrate our experience in ovarian torsion and provide an updated review of the available literature. Age at presentation, presenting symptoms, diagnostic studies, surgical procedure and pathological findings are illustrated.

Methods

We present a retrospective review of all patients (9-16 years old, Table 1) who presented to our institution with acute ovarian torsion from January 1st, 2008 to October 31st, 2014 and that were surgically managed in our hospital. Documentation regarding admission, radiology studies, laboratory results, and operative and pathology reports was reviewed. Age at operation and presenting symptoms were analyzed for each patient, as well as patient history and clinical examination. Trans-abdominal Ultrasonography (US) associated to Color Doppler Ultrasonography (CDU) was used in all patients, while for selected girls we also performed Computed Tomography (CT) and Magnetic Resonance Imaging (MRI) assays. We evaluated all patients in the emergency room for β-hCG and serum tumor markers (CA-125, α-FP, and CA-19.9). The nature of the masses - when present - was defined by histopathological features using histological analysis following the WHO classification.[25]

Table 1 Patients treated in our department for ovarian torsion

Age	9	10	11	12	13	14	15	16
No. of patients	4	1	5	6	5	1	5*	2
Period status	irregular	irregular	irregular	irregular	irregular	irregular	irregular	irregular
Menarche	absent	present	present	present	present	present	present	present

*One of these patients was treated twice for asynchronous recurrence.

Adopted surgical procedures included open surgery or laparoscopy, and were performed using either the conservative or the radical approach according to patient's condition. Adnexal conservation treatment was practiced as detorsion alone, or detorsion followed by subsequent surgical cystectomy, or tumorectomy with ovarian tissue preservation and possibly oophoropexy. We did not include in our cohort the cases involving neonatal patients.

Results

Over a 7 year period, 29 surgical interventions for ovarian torsions in 28 children and adolescents have been evaluated (1 patient presented asynchronous bilateral lesions) (Table 1). They represented the 1.5% of all non-neonatal abdominal emergencies in the same period. The mean age of these patients was 12.2 years (age range: 9-16 years). All patients arrived at our pediatric emergency unit for lower AAP lasting several hours, sometimes associated to vomit, and were submitted to clinical examination; diagnosis for ovarian torsion was obtained with trans-abdominal US, associated to CDU in the emergency room to allow differential diagnosis with other causes of AAP.[17]

US in most cases revealed an enlarged ovary with peripherally displaced follicles and hyperechoic central stroma. In 20/29 cases the ovary was dislocated in the midline; free pelvic fluid was seen in 22 cases. Doppler examination revealed reduced intra-ovarian venous flow in 20/29 cases, absent arterial flow in 3 cases, whirlpool sign of twisted vascular pedicle in 2 cases, while normal vascularity was found in 3 patients, probably related to the vascular supply from uterine arteries. In 20/29 cases US revealed the presence of an underlying ovarian lesion; particularly, we observed 12 cases presenting with an intraovarian anechoic cyst (mean size of the cyst: 3.6 cm, range: 2-7 cm), 9/12 uniloculated, 1/12 multiloculated and 2/12 with fluid-fluid level suggesting the presence of bloody components. In 5 cases the ovary had a complex appearance with a partially echogenic mass with posterior sound attenuation suggesting the presence of sebaceous material within the cyst cavity. Furthermore, a mural hyperechoic plug was found in 3 of these patients; these imaging findings suggested the correct final diagnosis of ovarian teratoma. Moreover, 2 patients presented a solid hypoechoic component within the cyst. One patient had a confirmed diagnosis of polycystic ovarian syndrome (PCOS), and both ovaries presented the typical hyperechoic central stroma with "string of pearl appearance follicles" of similar size. The controlateral ovary was normal in all other patients. In 3 patients it was necessary to perform a CT because of a suspected malignant mass, and only for one patient we also used MRI. The patient who underwent MRI presented an atypical clinical picture, and US initially showed a uterine mass; 24 hours after presentation, an ovarian right torsion was diagnosed and the patient was immediately submitted to surgical intervention. Interestingly, this patient presented 9 months later with a metachronous contralateral left ovary torsion.

We routinely evaluated quantitively both serum β-hCG and tumor markers (CA-125, α-FP, and CA-19.9) in the emergency room; we always found them within normal ranges. Ovarian torsion was located on the right side in 19 patients (66%), while only 10 times (34%) it involved the left ovary. Videolaparoscopy (VLP) was performed 11 times (38%), while the remaining 18 children were treated using a traditional open technique. Ovari-annessectomy was done 14 times (48%), simple ovariectomy 4 times (14%), cuneiform resection of the cyst 7 times, in 1 case we performed the excision of salpinx for a paraooforon cyst, and in 2 patients we made the derotation of annexes associated to oophorexy. In one of these last cases, the patient previously showed a metachronous adnexial torsion involving the right ovary, and in the first place she underwent an ovariectomy; on a later time, the same patient had a derotation of adnexa with prompt revascularization followed by left oophorexy. The histological examination after surgery revealed that the ovarian torsion occurred in an ovary with hemorrhagic cysts in 2 cases (7%), follicular cysts in 10 cases (34.5%), fibrous cystoadenoma in 2 cases (7%), mature cystic teratomas in 4 cases (14%), immature teratoma in 1 case (3.5%), and policystic ovary in 1 patient (3.5%). There were 9 torsions in 8 patients affecting normal ovaries, including one asynchronous recurrence of a left normal ovary 9 months after the involvement of the right one.

Discussion

Ovarian torsions represent an infrequent cause of AAP in children. The actual incidence of ovarian torsion in children is unknown.[18,26] Of all cases of ovarian torsion diagnosed, nearly three-fourths present during the reproductive years from 20 to 40 years of age, and a few occur in postmenopausal women.[18] The etiology, presentation, diagnosis and management of younger patients often differ from those of adults.[27] In children, the torsion of a normal ovary is more frequent than in adults because of the greater length of the ovarian pedicle.[28] An abnormally long fallopian tube, mesosalpinx or mesoovarium causes an excess of mobility of the adnexa, determining a higher risk of torsion in these girls.[29,30]

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As a general rule, right ovarian torsion is more frequent than that affecting the left one, because

i. The sigmoid colon reduces the chance of twisting,

ii. Of the hypermobility of the cecum and ileum and

iii. Of the slightly longer mesosalpinx and utero-ovarian ligament on the right, allowing more mobility of the adnexa.[31,32]

Clinical presentation is nonspecific, and diagnosis is based on a high index of suspicion. Ultrasound scan remains the most useful investigation; blood flow analysis through Doppler examination does not exclude ovarian torsion but it is useful to evaluate the blood flow in the affected ovary. Patients with adnexal torsion usually experience the sudden onset of intense pain.[33,34] However, this type of abrupt start is not typical and affects no more than half of the patients.[35] Therefore ovary torsion clinical presentation is unspecific and it can mimic many abdominopelvic surgical diseases - it may be confused for example with acute appendicitis, with which it shares also the possible presence of leucocytosis. The pain usually develops over a short period of time, from a few hours to less than two days from the beginning of the twist. Pain lasting more than 10 hours before surgery is associated with an increased rate of adnexal necrosis. The pain felt is usually very severe but not in every case.[36]

Ultrasonography has become a routinely used investigation method in our experience, this is in line with current clinical trends;[3] indeed, we carry it out already at check-in in the emergency room as part of the standard clinical examination. The procedure we apply involves an initial abdominal scan with the bladder full, to highlight any adnexal mass located high in the abdomen. We never performed transvaginal US. This US examination reveals a pathological adnexal image in most cases.[37,38] Warner and collaborators observed that the association of an adnexal mass (detected by US) and pain should prompt consideration of adnexal torsion as a primary diagnosis.[39] However, these abnormal adnexal images are not always pathognomonic of torsion and do not allow a differential diagnosis with other cystic pathologies. Similarly, between 9% and 26% of cases of torsion occur in apparently normal adnexa and consequently show no initial signs of abnormality at sonography.[37] In our experience, US was correct 28 times, and only one patient was requested for Magnetic Resonance Imaging (MRI).

Conclusion

Ovarian torsion in pediatric patients is frequently characterized by a different etiology, diagnosis and therapy if compared to its adult counterpart. In addition, it is an infrequent and unspecific cause of AAP in children. This might cause a delay in diagnosis and management, resulting in adnexal infarction and, in most extreme cases, loss of the ovary. It is thus pivotal to have a prompt diagnosis to preserve these structures. In case of AAP, we believe that in pediatric patients US is the most reliable and useful diagnostic tool, to be used in any case of abdominal pain presentation, and that it should be coupled with CDU for the correct evaluation of blood circulation in the suffering ovary.

Conflicts of Interest

The Authors declare that no conflicts of interest are present.

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References

1. Asfour V, Varma R, Menon P Clinical risk factors for ovarian torsion. *J Obstet Gynaecol*. 2015;35(7):721–725.
2. Spinelli C, Di Giacomo M, Cei M, Mucci N Functional ovarian lesions in children and adolescents: when to remove them. *Gynecol Endocrinol*. 2009;25(5):294–298.
3. Ashwal E, Krissi H, Hiersch L, et al. Presentation, diagnosis, and treatment of ovarian torsion in premenarchal girls. *J Pediatr Adolesc Gynecol*. 2015;28(6):526–529.
4. Houry D, Abbott JT Ovarian torsion: a fifteen–year review. *Ann Emerg Med*. 2001;38(2):156–159.
5. Servaes S, Zurakowski D, Laufer MR, Feins N, Chow JS Sonographic findings of ovarian torsion in children. *Pediatr Radio*. 2007;l 37(5) 446–451.
6. Adelman S, Benson CD, Hertzler JH. Surgical lesions of the ovary in infancy and childhood. *Surg Gynecol Obstet*. 1975;141(2):219–226.
7. Griffin D, Shiver SA. Unusual presentation of acute ovarian torsion in an adolescent. *Am J Emerg Med*. 2008;26(4):520.e1–520.e3.
8. Hoey BA, Stawicki SP, Hoff WS, et al. Ovarian torsion associated with appendicitis in a 5–year–old girl: a case report and review of the literature. *J Pediatr Surg*. 2005;40(9):E17–E20.
9. Anders JF, Powell EC. Urgency of evaluation and outcome of acute ovarian torsion in pediatric patients. *Arch Pediatr Adolesc Med*. 2005;159(6):532–535.
10. Huang TY, Lau BH, Lin LW, et al. Ovarian cyst torsion in a toddler. *Am J Emerg Med*. 2009;27(5):632.e1–632.e3.
11. Kloss BT, Prince LA. Ovarian torsion in a prepubertal girl. *Int J Emerg Med*. 2009;2(2):127–128.
12. Ryan MF, Desai BK. Ovarian torsion in a 5–year old: a case report and review. *Case Rep Emerg Med*. 2012;2012: 679121.
13. Cohen Z, Shinhar D, Kopernik G, et al. The laparoscopic approach to uterine adnexal torsion in childhood. *J Pediatr Surg*. 1996;31(11):1557–1559.
14. Guthrie BD, Adler MD, Powell EC. Incidence and trends of pediatric ovarian torsion hospitalizations in the United States, 2000–2006. *Pediatrics*. 2010;125(3):532–538.
15. Aziz D, Davis V, Allen L, et al. Ovarian torsion in children: is oophorectomy necessary? *J Pediatr Surg*. 2004;39(5):750–753.
16. Oltmann SC, Fischer A, Barber R, et al. Cannot exclude torsion – a 15–year review. *J Pediatr Surg*. 2009;44(6):1212–1216.
17. Rialon KL, Wolf S, Routh JC, et al. Diagnostic evaluation of ovarian torsion: an analysis of pediatric patients using the Nationwide Emergency Department Sample. *Am J Surg*. 2017;213(4):637–639.
18. Cass DL. Ovarian torsion. *Semin Pediatr Surg*. 2005;14(2):86–92.
19. Oelsner G, Shashar D. Adnexal torsion. *Clin Obstet Gynecol*. 2006;49(3):459–463.
20. Laganà AS, Sofo V, Salmeri FM, et al. Oxidative stress during ovarian torsion in pediatric and adolescent patients: changing the perspective of the disease. *Int J Fertil Steril*. 2016;9(4):416–423.
21. Smorgick N, Melcer Y, Sarig–Meth T, et al. High risk of recurrent torsion in premenarchal girls with torsion of normal adnexa. *Fertil Steril*. 2016;105(6):1561–1565.e3.
22. Karaman E, Beger B, Çetin O, et al. Ovarian torsion in the normal ovary: a diagnostic challenge in postmenarchal adolescent girls in the emergency department. *Med Sci Monit*. 2017;23:1312–1316.
23. Tintinalli J. Emergency Medicine: a comprehensive study guide (McGraw Hill Professional) pp. 2004;904.
24. Bar–On S, Mashiach R, Stockheim D, et al. Emergency laparoscopy for suspected ovarian torsion: are we too hasty to operate? *Fertil Steril*. 2010;93(6):2012–2015.

25. Tavassoli FA, Devilee P. World Health Organization classification of tumours. Pathology and genetics of tumours of the breast and female genital organs. IARC Press. Lyon, France. 2003.
26. Beaunoyer M, Chapdelaine J, Bouchard S, et al. Asynchronous bilateral ovarian torsion. *J Pediatr Surg*. 2004;39(5):746–749.
27. Nur Azurah AG, Zainol ZW, Zainuddin AA, et al. Update on the management of ovarian torsion in children and adolescents. *World J Pediatr*. 2015;11(1):35–40.
28. Childress KJ, Dietrich JE. Pediatric ovarian torsion. *Surg Clin North Am*. 2017;97(1):209–221.
29. Kamio M, Oki T, Inomoto Y, et al. Torsion of the normal ovary and oviduct in a pre–pubertal girl. *J Obstet Gynaecol Res*. 2007;33(1):87–90.
30. Huchon C, Fauconnier A. Adnexal torsion: a literature review. *Eur J Obstet Gynecol Reprod Biol*. 2010;150(1):8–12.
31. Blitz MJ, Appelbaum H. Management of isolated tubal torsion in a premenarchal adolescent female with prior oophoropexy: a case report and review of the literature. *J Pediatr Adolesc Gynecol*. 2013;26(4):e95–e97.
32. Mellor A, Grover S. Auto–amputation of the ovary and fallopian tube in a child. *Aust N Z J Obstet Gynaecol*. 2014;54(2):189–190.
33. Nichols DH, Julian PJ. Torsion of the adnexa. *Clin Obst Gynecol*. 1985;28(2):375–380.
34. Goldstein DP. Acute and chronic pelvic pain. *Pediatr Clin N Am*. 1989;36(3):573–580.
35. Lomano JM, Trelford JD, Ullery JC. Torsion of the uterine adnexa causing an acute abdomen. *Obstet Gynecol*. 1970;35(2):221–225.
36. Abbott J. Pelvic pain: lesson from anatomy and physiology. *J Emerg Med*. 1990;8(4):441–447.
37. Bider D, Mashiah S, Mordechai D, et al. Clinical, surgical and pathologic findings of adnexal torsion in pregnant and non pregnant women. *Surg Gynecol Obstet*. 1991;173(5):363–365.
38. Chang HC, Bhatt S, Dogra VS. Pearls and pitfalls in diagnosis of ovarian torsion. *Radiographics.* 2008;28(5):1355–1368.
39. Warner MA, Fleischer AC, Edell SL, et al. Uterine adnexal torsion: sonographic findings. *Radiology*. 1985;154(3):773–775.

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