

Case Report

Bilateral Paraovarian Cysts with Torsion in Children: A Case Report and Literature Review

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Paraovarian cysts are located in the broad ligament between the ovary and the fallopian tube. They rarely cause symptoms and occasionally are incidentally found. Symptoms occur when they develop complications such as torsion. Clinical torsion of a paraovarian cyst is uncommon in the pediatric age group and difficult to distinguish from other causes of acute abdominal pain.

We present a 12-year-old girl who had bilateral paraovarian cysts, which torsion of the right side caused acute abdominal pain mimicking the presentation of acute appendicitis.

A certain pre-operative diagnosis of torsion paraovarian cyst in children with abdominal pain is challenging. Careful history taking, physical examination, investigation and consistent re-evaluation of abdominal signs may facilitate appropriate decision making before surgery.

Keywords: Paraovarian cyst, Paratubal cyst, Hydatid of Morgagni, Childhood

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The incidence of paraovarian cyst is about 7 to 10%⁽¹⁾ of all adnexal masses. Clinical torsion of a paraovarian cyst is uncommon and difficult to distinguish from other causes of acute abdominal pain. Of all the adnexal causes of abdominal pain, torsion of ovarian masses is the most common. Isolated torsion of the fallopian tube has also been reported, however, torsion of the paraovarian cyst is rare, especially in children. We present a case of bilateral paraovarian cysts which torsion on the right side caused acute abdominal pain mimicking appendicitis.

Case Report

An otherwise healthy 12-year-old girl presented with right lower abdominal pain for 12 hours. Although she had no fever, diarrhea or urinary tract symptoms, she complained of being nauseous and vomited some ingested food since the beginning of her symptoms. Her past history consisted of regular menstruation, no abnormal discharge, no underlying disease, no current medication and no history of abdominal trauma. Physical examination showed a body

temperature of 36.0°C, pulse rate of 78 per minutes and blood pressure was 142/90 mmHg. The patient's height was 160 cm and weight was 74 kg. Her abdomen was soft with point of maximal tenderness at right lower quadrant (RLQ). She had hypoactive bowel sound and no rebound tenderness nor palpable mass at initial physical examination. Per rectal examination demonstrated normal findings with some retained fecal material. Laboratory studies showed Hb 13.2 g/dl, Hct. 42%, Plt. 270,000 and WBC 11,600 (N 73, L29). Other blood chemistries were within normal range. Plain abdominal x-ray showed normal bowel gas pattern and some fecal impaction in the left side colon. The patient was managed as constipation with fleet enema.

After several hours of close abdominal sign observation, she developed fever of 37.8°C and complained of increase tenderness at RLQ area. Re-evaluation of abdominal signs detected localizing tenderness at RLQ and rebound tenderness positive. An emergency appendectomy was scheduled. After general anesthesia was administered, we re-evaluated the abdomen and a palpable tense cystic mass at suprapubic area was found. The mass persisted in the suprapubic area regardless of the urinary catheter being retained. Pre-operative ultrasonography was performed and revealed a cystic mass at lower abdomen superior to the urinary bladder (Fig. 1), the origin of the cystic mass could not be clearly identified. A diagnosis of

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torsion of the ovarian cyst was suspected and laparotomy was performed via a low midline incision.

Intra-operative findings consisted of approximately 30 ml of serosanguinous fluid in pelvic cavity, twisted right paraovarian cyst, size 8.0x10.0 cm in diameter, with hemorrhagic fluid content. The right ovary adhered to this paraovarian cyst had signs of inflammation and was actively bleeding (Fig. 2). There were multiple, varying in size, paraovarian cysts in the left adnexal area without complications. The uterus was normal size and the left ovary and fallopian tube were unremarkable (Fig. 3). Right salpingo-oophorectomy and excision of left paraovarian cyst was performed.

After the operation, the patient had an uneventful post-operative recovery and was discharged from the hospital on the 3rd post-operative day. Pathologic examination demonstrated benign serous lining of the cyst wall corresponding with simple cysts, consistent with paraovarian cysts on both sides. The tumor markers taken subsequently were within normal limits (AFP 1.10 IU/ml, BHCG <0.1 mIU/ml, CEA 1.01 ng/ml).

Discussion

Paraovarian cysts are simple cysts located in the broad ligament between the ovary and the fallopian tube and constitute about 10% of adnexal masses⁽²⁾. They originate from the tissue of the broad ligament, predominantly from mesothelium covering the peritoneum and also from paramesonephric (Mullerian duct) and mesonephric (Wolffian duct) remnants. The size of paraovarian cysts vary, ranging from 1 to 10 cm

in diameter. Occasionally, it can become very large most likely from hormonal effect. When a paraovarian cyst is pedunculated and located near the fimbria of the fallopian tube, it is referred to as an hydatid cyst of Morgagni, which is usually smaller than 2 cm. In the literature, paraovarian cyst and paratubal cyst are usually used synonymously.

The true incidence of paraovarian cyst in children is unclear. There was a study reporting the incidence of paraovarian cysts among benign adnexal

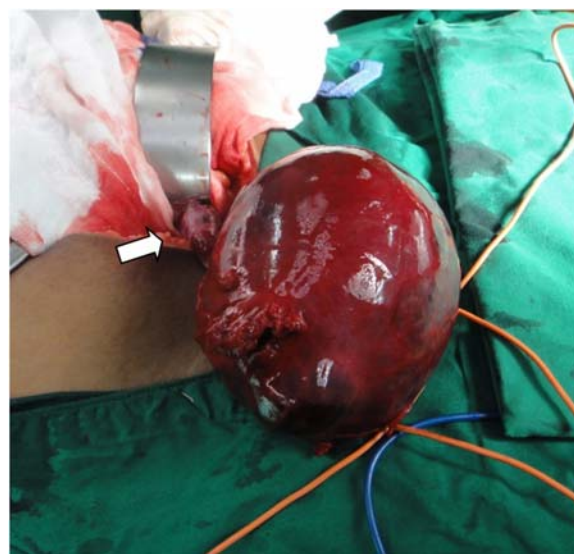


Fig. 2 Twisted right paraovarian cyst, size 8.0x10.0 cm (white arrow showed right ovary).



Fig. 1 Pre-operative ultrasonography revealed cystic mass at lower abdomen superior to the urinary bladder, size 8.0x10.0 cm.

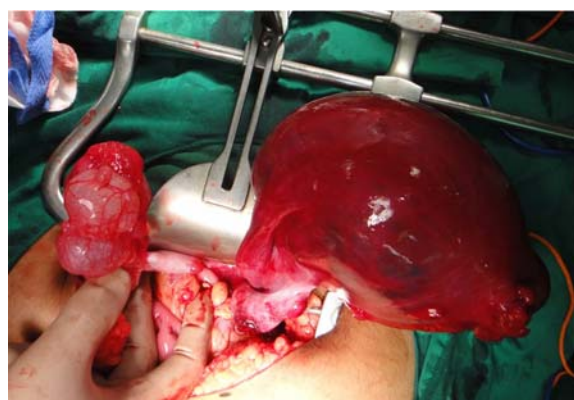


Fig. 3 The large right paraovarian cyst, after detorsion and the smaller left multiple, varying in size, paraovarian cysts without complication, the left ovary was normal.

cysts to be between 17% and 33% involving both children and adults⁽²⁻⁵⁾. There have been reports in all age groups, ranging from pre-menarchial period up to menopause. Paraovarian cysts have been reported more common in reproductive age women between 30 and 40 years of age or during pregnancy, although very uncommon in children⁽⁶⁾. However in a recent study, a much higher incidence of 7.3% paraovarian cyst was reported in pediatric and adolescent population⁽¹⁾.

A certain pre-operative diagnosis of paraovarian cyst is very difficult. Therefore, they are usually found incidentally during operative procedures for other indications. Barloon et al concluded that these cysts are difficult to diagnose prior to surgery despite using ultrasonography and pre-operative diagnosis was only possible in 6.6% of the cases⁽²⁾. A pre-operative misdiagnosis as true ovarian cysts is very common. However, a paraovarian cyst should be suspected if the ipsilateral normal ovary was identified, and the cyst was located near the uterus.

Although they are uncommon and usually incidentally found, they rarely cause any symptoms. Symptomatic paraovarian cysts share a common clinical presentation of acute abdomen as well as other diseases such as acute appendicitis, torsion or ruptured ovarian cyst, acute ureteric colic, or PID. Clinical torsion of paraovarian cyst is actually uncommon. The incidence of torsion among patients with paraovarian cyst was 2.1% to 16%, which is comparable to an incidence of 2.3% among 215 patients with adnexal cysts during the same period⁽⁴⁾. Conversely, in children, torsion is a more common issue when the infundibulo-pelvic ligament is longer⁽⁶⁾. In young females, operated for RLQ abdominal pain, an incidence of 13.6% had paraovarian cyst and two of which had isolated torsion of a paraovarian cyst⁽⁷⁾. Since 1997, there were 12 cases of torsion paraovarian cyst in children reported in the English literature (Table 1). The case reported here as torsion paraovarian cyst is the 13th^(3,8-15).

Regarding the diagnosis of a complicated paraovarian cyst such as torsion, the physical findings mostly included abdominal pain and adnexal tenderness on pelvic examination. Peritoneal irritation signs and sometimes a palpable mass may or may not be acquired from general physical examination. Laboratory values are usually non-specific. Therefore a torsion paraovarian cyst, although rare, should be kept in mind when differential diagnosing the cause of acute abdominal pain in children. The ultrasonography for the diagnosis of paraovarian cyst, is a simple, non-invasive yet useful investigation, however, it is operator

dependent. Other imaging techniques, such as CT scan and MRI can also be used in differential diagnosis. There is no specific finding on non-contrast CT scan other than unilocular cystic mass near the adjacent ovary. MRI features of paraovarian cyst however can demonstrate a normal ovary separating it from the cyst. Surgical management of twisted paraovarian cysts has been reported in both laparoscopic and laparotomy approach. Cystectomy is recommended and ovarian salvage should be considered as priority. The ovary should not be resected if the blood supply to the ovary could be preserved in a twisted paraovarian cyst. Internal drainage procedures, such as laparoscopic unroofing and needle aspiration, also have been used for physiologic ovarian cyst as minimally invasive treatments. However, we consider that these procedures are not suitable for paraovarian cyst on account of the risk for malignant change^(3,5).

Regarding histopathology, paraovarian cysts are usually simple cysts filled with serous fluid. Although rare, paraovarian cyst containing neoplasm has been reported⁽⁵⁾. If papillary projections were present, the risk of malignancy increases. Concerning the origin, Genadry et al analyzed 132 paraovarian cysts and found that the majority of 68% were classified as mesothelial, 30% as paramesonephric and only 2% originated from mesonephric remnants⁽⁴⁾.

From our experience in this case, the patient presented with complication of a twisted right paraovarian cyst with clinical signs mimicking acute appendicitis. No intra-abdominal mass could be palpated owing to the limitation of thick-bodied abdominal wall. Subsequently, after general anesthesia was administered, we re-examined her abdomen and the pelvic mass could be palpated. The ultrasonography was very useful in determining the nature of the mass, a non-invasive and good modality for pre-operative evaluation, which could be performed at bed side. According to the ultrasonography findings, the more common, twisted ovarian tumor was suspected initially. A lower midline incision was performed for proper good exposure. Accordingly, the gangrene twisted right paraovarian cyst and profoundly bleeding right ovary were found. Therefore, a right salpingo-oophorectomy was inevitable. The normal left ovary was preserved and the non-complicated left paraovarian cyst removed.

Conclusion

A certain pre-operative diagnosis of torsion paraovarian cyst in children with abdominal pain is challenging. Careful history taking, physical

Table 1. Reported paraovarian cysts with torsion in children in the English literature

Year	Author (ref)	Age (years)	Presentation	Imaging results	Indication for surgery	Surgical procedure	Pathology
1997	Berlin SC et al ⁽⁸⁾	10	POC torsion	US: Rt. cystic mass 4.0x4.7	Acute appendicitis	Laparotomy with Rt. SO	No report
2001	Lurie S et al ⁽⁹⁾	12.5	Torsion Rt. POC	CT: twisted Rt. adnexal mass 6 cm	Torsion of fallopian tube	Laparoscopic detorsion and cystectomy	Benign POC
2002	Okada et al ⁽³⁾	14	POC torsion	CT: twisted Rt. fallopian tube 10x12 cm	Torsion of fallopian tube	Laparotomy with Rt. SO	Benign POC
2007	Szczepanik et al ⁽¹⁰⁾	14	Torsion Lt. POC	CT: homogeneous cyst close to the uterus 5.5x7.5 cm	Torsion of adnexal cyst	Laparotomy with cystectomy	Benign POC
2007	Szczepanik et al ⁽¹⁰⁾	15	Torsion Rt. POC	CT: Rt. side non-ovarian mass 6 cm	Torsion Rt. POC	Laparotomy with cystectomy	Benign POC
2008	Kostov M, et al ⁽¹¹⁾	14	Bilateral adnexal torsion	CT: Large hypodensity mass 30x26x12 occupying entire abdomen	Torsion of adnexal mass	Laparotomy with cystectomy	Benign serous cystadenoma
2011	Muthucumaru M, et al ⁽¹²⁾	14	Worsening RLQ pain	US: Bilateral polycystic ovaries, free fluid in pouch of Douglas	Torsion Rt. fimbrial Hydatid of Morgagni cyst	Laparoscopic detorsion and cyst removal	Hemorrhagic infarction of the cyst
2011	Kitporntheranunt M, et al ⁽¹³⁾	11	Sudden RLQ pain	Diag lap: normal appendix, hemorrhage and torsion right hydatid of Morgagni cyst	Torsion of Rt. fimbrial Hydatid of Morgagni	Laparoscopic detorsion, cyst excision and appendectomy	Hemorrhagic infarction lined with ciliated columnar cells
2011	Kitporntheranunt M, et al ⁽¹³⁾	11	Bilateral torsion adnexal mass	CT: Bilobulated cystic mass 10x12x12 in left adnexa, another 10x6x5 in pelvis	Entangled bilateral adnexal torsion	Laparoscopic detorsion and bilateral ovarian cystectomy	Benign POC
2012	Erdal T, et al ⁽¹⁴⁾	12	Torsion Rt. POC	US: cystic mass 7.7x8.2	Rt. ovarian torsion	Laparotomy with Rt. SO	Mesonephric POC
2013	Yilmaz Y et al ⁽¹⁵⁾	11	Lt. POC cyst	US: simple cyst 5.5x3.3 cm close to Lt. ovary	Twisted Lt. adnexal mass	Laparotomy with cystectomy	Benign POC
2013	Case	9	Rt. adnexal mass	US: Solid cystic mass 5 cm in lower abdomen	Twisted Rt. ovary and Left adnexal mass	Laparotomy detorsion and cystectomy	Benign POC
2013	Case	12	Acute RLQ pain	US: Cystic mass 8.0x10.0 cm at Rt. adnexa	Suspected twisted Rt. ovarian tumor	Laparotomy with Rt. SO and excision Lt. POC	Benign POC

POC = paraovarian cyst; US = ultrasonography; TVS = transvaginal US; CT = computed tomography; SO = salpingo-oophorectomy

examination, appropriate investigation and consistent re-evaluation of abdominal signs may facilitate appropriate decision-making in choosing the best surgical incision and procedure for each patient.

What is already known on this topic?

Paraovarian cysts are rare in pediatric population. It is more common in female of reproductive age group and can sometimes be a cause of surgical emergency, often times indistinguishable from ovarian emergencies.

What this study adds?

The objective of this article is to raise awareness in approaching acute lower abdominal pain in female pediatric population, which the condition could be the cause and an appropriate investigation should be considered prior to surgical procedure. The correct diagnosis can determine the surgical incision and procedure that is best for the patient.

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Potential conflicts of interest

None.

References

1. Muolokwu E, Sanchez J, Bercaw JL, Sangi-Haghpeykar H, Banszek T, Brandt ML, et al. The incidence and surgical management of paratubal cysts in a pediatric and adolescent population. *J Pediatr Surg* 2011; 46: 2161-3.
2. Barloon TJ, Brown BP, Abu-Yousef MM, Warnock NG. Paraovarian and paratubal cysts: preoperative diagnosis using transabdominal and transvaginalsonography. *J Clin Ultrasound* 1996; 24: 117-22.
3. Okada T, Yoshida H, Matsunaga T, Kouchi K, Ohtsuka Y, Takano H, et al. Paraovarian cyst with torsion in children. *J Pediatr Surg* 2002; 37: 937-40.
4. Genadry R, Parmley T, Woodruff JD. The origin and clinical behavior of the parovarian tumor. *Am J Obstet Gynecol* 1977; 129: 873-80.
5. Smorgick N, Herman A, Schneider D, Halperin R, Pansky M. Paraovarian cysts of neoplastic origin are underreported. *JSL* 2009; 13: 22-6.
6. Kiseli M, Caglar GS, Cengiz SD, Karadag D, Yilmaz MB. Clinical diagnosis and complications of paratubal cysts: review of the literature and report of uncommon presentations. *Arch Gynecol Obstet* 2012; 285: 1563-9.
7. Vlahakis-Miliaras E, Miliaras D, Koutsoumis G, Miliaras S, Spyridakis I, Papadopoulos MS. Paratubal cysts in young females as an incidental finding in laparotomies performed for right lower quadrant abdominal pain. *Pediatr Surg Int* 1998; 13: 141-2.
8. Berlin SC, Morrison SC, Myers MT, Sivit SJ, Weinert DM. Pediatric case of the day. Parovarian cysts. *AJR Am J Roentgenol* 1997; 169: 306, 308-6, 310.
9. Lurie S, Golan A, Glezerman M. Adnexal torsion with a paraovarian cyst in a teenage girl. *J Am Assoc Gynecol Laparosc* 2001; 8: 597-9.
10. Szczepanik ME, Wittich AC. Para-ovarian cyst torsion mimicking an acute abdomen in adolescent. *J Gynecol Surg* 2007; 23: 63-8.
11. Kostov M, Mijovic Z, Mihailovic D. Giant paraovarian cyst in a child complicated with torsion. *Vojnosanit Pregl* 2008; 65: 843-6.
12. Muthucumar M, Yahya Z, Ferguson P, Cheng W. Torsion of hydatids of Morgagni in premenarchal adolescent girls—a case report and review of literature. *J Pediatr Surg* 2011; 46: e13-e15.
13. Kitporntheranunt M, Wong J, Siow A. Entangled bilateral adnexal torsion in a premenarchal girl: a laparoscopic approach. *Singapore Med J* 2011; 52: e124-e127.
14. Turk E, Karaca F, Tan A, Yilmaz UM. Giant paraovarian cysts in children: two different ages and clinical pictures. *J Clin Anal Med* 2015; 6: 369-71.
15. Yilmaz Y, Ozen IO, Caliskan D, Dilmen U. Paraovarian cyst torsion in children: report of two cases. *PediatrInt* 2013; 55: 795-7.

ภาวะถุงน้ำที่พังผืดยึดท่อนำไข่ทั้งสองข้างในผู้ป่วยเด็ก: รายงานผู้ป่วยและบททวนวรรณกรรม

ทิภาพร ทับทอง, มงคล เลหาพิณแสง

ภาวะถุงน้ำที่เยื่อพังผืดยึดท่อนำไข่หรือปีกมดลูก (Paraovarian cyst) เป็นภาวะที่พบบ่อยในกลุ่มผู้ป่วยเด็ก หากแต่พบบ่อยในหญิงวัยเจริญพันธุ์เนื่องจากเชื่อว่าเป็นที่เกิดจากฮอริโมน โดยปกติถุงน้ำนี้จะมีอาการจนกว่าเกิดภาวะแทรกซ้อนเช่นการบิดหรือตกเลือดหรือขนาดโตขึ้นเป็นเนื้องอก จะทำให้ผู้ป่วยมีอาการปวดท้องน้อย เป็นๆ หายๆ หรือทวีความรุนแรงจนเกิดภาวะฉุกเฉินทางศัลยกรรมได้

ผู้เขียนได้รายงานผู้ป่วยเด็กอายุ 12 ปีที่มาพบแพทย์ด้วยอาการปวดท้องน้อยข้างขวาเป็นๆ หายๆ และต่อมาอาการปวดมีความรุนแรงมากขึ้นร่วมกับภาวะคลื่นไส้อาเจียน เบื่ออาหาร มีไข้ จากการตรวจร่างกาย และตรวจทางห้องปฏิบัติการเบื้องต้นสงสัยภาวะไส้ติ่งอักเสบ จนกระทั่งไปห้องผ่าตัดแพทย์ตรวจร่างกายซ้ำ และยืนยันด้วยการตรวจอัลตราซาวด์จึงพบถุงน้ำที่บริเวณข้างรังไข่ ช่วยให้แพทย์ตัดสินใจลงแผลผ่าตัดตรงกลาง พบว่ามีถุงน้ำที่พังผืดยึดท่อนำไข่ทั้งสองข้างโดยข้างขวาบิดและขาดเลือด จำเป็นต้องตัดปีกมดลูกและรังไข่ข้างนั้นออกพร้อมกับเอาถุงน้ำข้างซ้าย และเก็บรังไข่ข้างซ้ายไว้ได้ จากการทบทวนวรรณกรรมพบว่ามีการรายงานภาวะถุงน้ำที่พังผืดยึดท่อนำไข่ในเด็กไม่มาก

ภาวะถุงน้ำที่พังผืดยึดท่อนำไข่พบบ่อยในผู้ป่วยเด็ก การวินิจฉัยก่อนผ่าตัดทำได้ยาก การซักประวัติ ตรวจร่างกายและตรวจเพิ่มเติมที่แม่นยำ ตลอดจนการติดตามอาการของผู้ป่วยอย่างใกล้ชิดจะช่วยให้ผ่าตัดรักษาได้อย่างเหมาะสม
