

A voluminous twisted paraovarian cyst in a 74-year-old patient: case report and review of the literature

**M. Varras¹, M.D., Ph.D.; Ch. Akrivis², M.D., Ph.D.; D. Polyzos¹, M.D.; S. Frakala¹, M.D.;
Ch. Samara³, M.D., Ph.D.**

¹*Department of Gynaecology, "George Gennimatas" General State Hospital, Athens,
Second District National Health System, Athens*

²*Department of Obstetrics and Gynaecology, "G. Chatzikosta" General State Hospital,
Ioannina, District National Health System of Epirus*

³*Department of Ultrasound, Computed Tomography, MRI, "George Gennimatas" General State Hospital, Athens,
Second District National Health System, Athens (Greece)*

Summary

Although paraovarian cysts rarely cause symptoms, they may be complicated due to massive size, torsion or internal haemorrhage from rupture. Moreover, benign or malignant neoplasms may occasionally develop in paraovarian cysts. The risks from voluminous ovarian or paraovarian cysts are due to severe cardiovascular, pulmonary, and circulatory problems including surgical and postoperative complications. We present a case of a 74-year-old patient who suffered from a twisted voluminous right paraovarian cyst. Her preoperative respiratory function required attention. The patient was treated surgically with good results and she showed notable improvement of her respiratory function, postoperatively. The maximum diameter of the mass was 26 cm and its total weight was 5,100 g. In addition, a simple cyst was found in the left ovary with a maximum diameter of 9.5 cm and total weight of 300 g. In conclusion, paraovarian cysts, even in elderly patients, can reach large sizes requiring awareness of the problems that these large masses may cause.

Key words: Paraovarian cyst; Complications; Torsion; Twisted; Computed tomography; Voluminous paraovarian tumor.

Introduction

Paraovarian cysts constitute 10-20% of all adnexal tumor-like lesions in pathological series [1-3], which are considered to be either of mesonephric, mesothelial, or paramesonephric origin [1, 2, 4-6]. They occur over a wide age range with a peak incidence in the fourth and fifth decades of life [2, 6, 7]. Paraovarian cysts are often discovered incidentally during surgery usually performed for other pathologic conditions [1]. Paraovarian cysts have the same complications that are encountered with ovarian cystic lesions, such as torsion or internal haemorrhage from rupture [6, 8]. There are also scattered reports of benign and malignant neoplasms developing in paraovarian cysts [8-12]. Paraovarian cysts have been reported to grow rapidly during pregnancy [1, 7], but one case of a paraovarian cyst unchanged throughout pregnancy has also been reported [13].

Voluminous intra-abdominal tumors change the anatomy and physiology of the patient and their excision may be associated with considerable mortality [14]. The increased size of the tumor can cause pulmonary effects in the patient due to a marked decrease in lung volume, pronounced rib flaring and attenuated diaphragmatic and abdominal muscles [14]. Operative problems include the risks of

anaesthesia, blood loss and hypothermia during a long operative period [14]. Postoperatively, cardiopulmonary dysfunction may result from several factors [15-17].

In this study, an unusual case of a voluminous twisted paraovarian cyst is presented together with the clinical, computed tomography scan and surgical findings. The problems associated with enormous paraovarian cysts are also discussed and the international literature is reviewed.

Case Report

A 74-year-old, nulliparous woman presented at the emergency department with diffused abdominal pain and vomiting during the previous three days prior to admission. The pain was more intense in the lower abdomen. The patient had a medical history of chronic obstructing pneumonopathy and ischemic heart disease. An operation for appendectomy was performed 50 years before. She suffered from thyroid disease and was treated with sodium levothyroxine. Physical examination on admission revealed abdominal tenderness that was more intense in the lower abdomen. Rebound tenderness was also found, which was becoming localized in the right iliac fossa. In addition, there was cystic abdominopelvic swelling. On palpation, the top of the mass was found to expand to the left costal arch. Peristalsis was absent. A pelvic examination revealed a very painful movement of the cervix; the uterus and adnexa were not distinguishable because of the huge abdominal-pelvic mass. Her temperature was 37.8°C. Initial laboratory studies showed a Hb of 14.2 g/dl; Ht 40.3%; WBC, 19.4 x 10³/µl with 83.1%

Revised manuscript accepted for publication March 3, 2003

polymorphonuclear leukocytes. The erythrocyte sedimentation rate was 46 mm/h. Arterial blood gas analyses showed a pH of 7.56, $p\text{CO}_2$ 40 mm Hg and $p\text{O}_2$ 62.3 mm Hg. Admission total protein, albumin, clotting studies, serum ammonia, urea and creatinine were normal. Liver function tests were unremarkable apart from total bilirubin, 1.8 mg/dl (normal values between 0.2 and 1 mg/dl) and conjugated bilirubin, 0.7 mg/dl (normal values between 0.1 and 0.5 mg/dl). Serum tumor makers were normal.

Chest X-ray showed an elevated left hemi-diaphragm. The abdominal X-rays showed huge shading with clear upper borders which expelled the *intestinum crassum* and the *intestinum tenue* towards the circumference and above. An abdominal/pelvic computerized tomography scan demonstrated a large bilocular cystic mass (Figure 1), which expanded from the level of the internal genital organs in contact with the uterus to the mesocolon. The mass had clear margins and a thin wall (Figure 2). Based on this the presence of an ovarian cystadenoma was indicated.

Internal medicine, cardiology, and anaesthesia consultations were obtained preoperatively to assess the patient's status. Incentive spirometry and deep breathing were taught to the patient and she had vigorous pulmonary toilet. The patient was also treated with oxygen therapy and bronchodilators. Her respiratory condition improved and an exploratory laparotomy was



Figure 1. — Computed tomography scan showing the presence of a large bilocular cystic mass and giving the appearance of an ovarian cystadenoma.

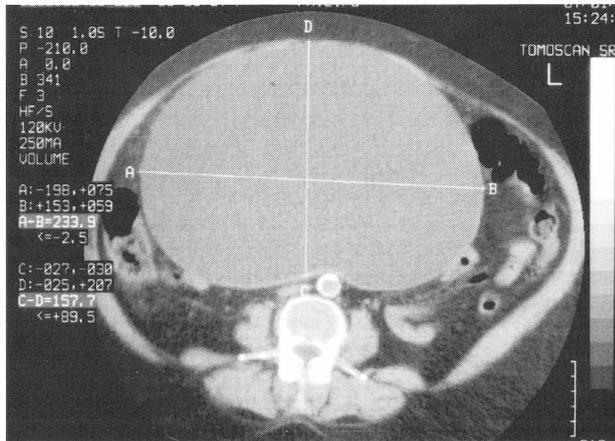


Figure 2. — Computed tomography scan showing the clear margins and thin wall of the mass, which subsequently was demonstrated to be a voluminous twisted right paraovarian cyst.

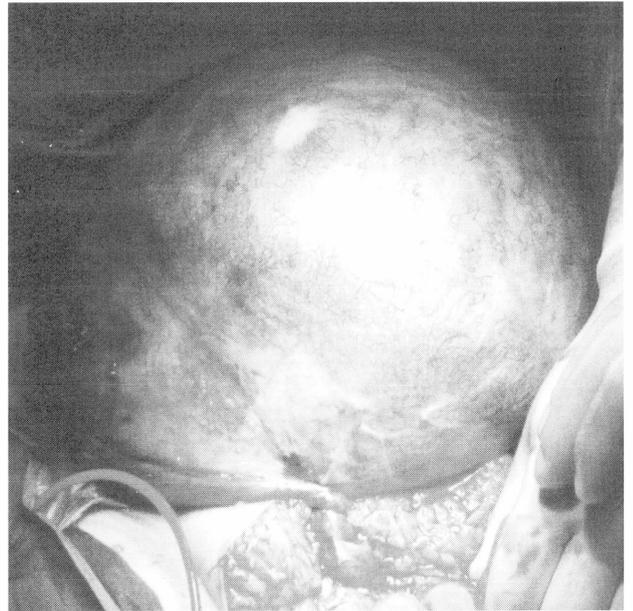


Figure 3. — The right paraovarian cyst was twisted clockwise around its peduncle.

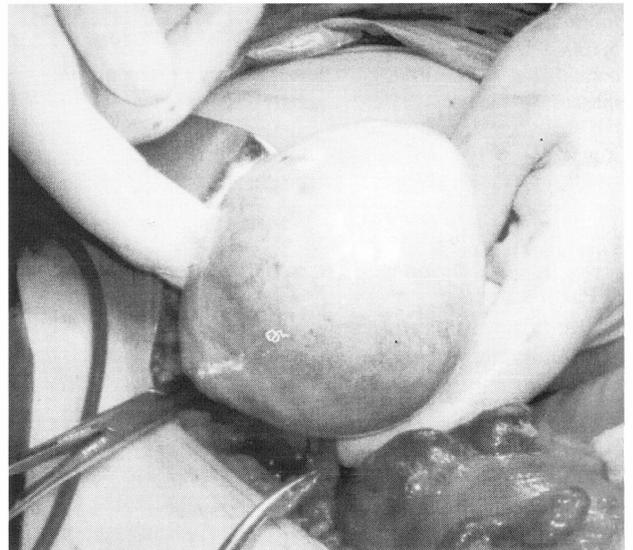


Figure 4. — A second cystic mass was found in the left ovary. The pathological examination showed that this mass was a simple ovarian cyst.

performed three days after admission. Crystalloid infusion was begun in order to optimize intravascular volume and cardiac output. Induction of anaesthesia was performed in reverse Trendelenburg in order to decrease pressure on the lungs and improve oxygenation. A wide ellipse of skin and subcutaneous tissue from the xyphoid to the pubic symphysis, along the medial margins of the separated recti was performed according to the operative technique described by Ferguson [18]. A voluminous pelvic mass was confirmed on the right. The mass was twisted clockwise through complete turns around its peduncle (Figure 3). The maximum diameter of the mass was 26 cm. Also in the left ovary a cystic mass was found measuring 9.5 x 8.5 x 9 cm (Figure 4). The uterus was of normal size for the patient's age and both ovaries and fallopian tubes were normal. Subtotal

abdominal hysterectomy with bilateral salpingo-oophorectomy was performed. The patient's vital signs and haemodynamic parameters remained stable throughout the procedure. During the closure of the abdominal wall the peritoneum, fascia and recti were brought together in the midline and overlapped slightly using a double absorbable suture. The patient was taken to the intensive care unit for one day and she was extubated on the day of operation. A liquid diet was begun on the third post-operative day, with return of bowel function. The postoperative course was uneventful, while with the resection of the intra-abdominal masses the patient showed notable improvement of the respiratory function. She was discharged on postoperative day 10.

The pathology report demonstrated the presence of a unilocular twisted paraovarian cyst on the right, weighing 5,100 g, which was filled with a pale yellowish fluid. Ovarian tissue from the right was not found. The mass of the left ovary was a simple ovarian cyst weighing 300 g. The body of the uterus and the fallopian tubes were without significant findings. Peritoneal washings were negative for malignancy.

Discussion

Examination of the primordial of the male and female internal genitalia showed that there are two separate sets of primordial, each of which is unipotential [19]. These are termed Wolffian or mesonephric ducts (male) and Müllerian or paramesonephric ducts (female) [19]. In the normal male the androgens secreted in considerable amounts by the testes actively induce the Wolffian ducts in the fourth week of the embryo stage to develop and give rise to epididymis, vas deferens and seminal vesicles [19]. In the female, the Wolffian ducts regress normally [19] and the paramesonephric ducts (Müllerian duct) give rise to the uterus, tubes and upper part of the vagina in the ninth week of gestation [19, 20]. The mesothelial epithelium covers the whole of the celomatic embryo cavity and forms the parietal and visceral peritoneum in the abdominal cavity [20]. Some residue on the broad ligament of mesonephric, paramesonephric and mesothelial tissue may cause paraovarian cysts [13, 20].

The terminology involving paraovarian cysts is highly individual and confusing; references to paratubal cysts, Wolffian cysts, fimbrial cysts, hydatid cysts of Morgagni and cysts of the organ of Rosenmüller can be found in the international literature [1, 7, 13]. However, the classification of paraovarian cysts has been simplified, depending on their origin, into the following basic types: (i) paramesonephric type, (ii) mesothelial type, (iii) mesonephric type [1, 2, 4, 5]. Genadry *et al.*, analysed 132 paraovarian cysts and found that most (68%) were classified as mesothelial; 30% as paramesonephric and only 2% arose from mesonephric elements [6]. Samaha *et al.*, described in a study of 79 paraovarian lesions that 60 were paramesonephric (75%), 19 mesothelial (24%) and one mesonephric (1%) [5]. The paramesonephric type paraovarian cysts are lined by a single-layered cuboidal or columnar epithelium. Some cells have a ciliary surface. Mucous secretion is occasionally present with mucous droplets in the cytoplasm. This lining epithelium is

similar to tubal epithelium and shows the same response to sex hormones as does tubal epithelium. Also, the surface structure of paramesonephric type paraovarian cysts is similar to the epithelium of ovarian surface tumors [1, 4, 7, 21]. A fine layer of connective tissue stroma with an admixture of smooth muscle, collagen and elastic fibers form the wall of the cyst [7]. During the development of the fetus multiple invaginations near the ostium of the fallopian tube become the fimbriae. Any secondary invagination that does not connect may form a blind sac, which may enlarge, thus forming a paraovarian cyst. Commonly these cysts are referred to as hydatid cysts of Morgagni and are usually small and often pedunculated [13]. The mesothelial type paraovarian cysts have a flat epithelium and are thought to represent peritoneal inclusion cysts arising from the mesothelium covering the adjacent peritoneum [1, 5, 6, 13]. The lining epithelium of the mesonephric type paraovarian cysts originate in a single layer of non-ciliated cuboidal or flattened epithelium [1, 6, 7]. The characteristics of the cysts of mesonephric origin are the low cuboidal non-secretory epithelium with its relatively thick muscular layer [7]. The stroma consists of dense connective tissue with both smooth muscle and fiber [7]. The mesonephric tubules are embryologic remnants located in the hilus of the ovary. Near the fallopian tube, the mesonephric tubules coalesce to join the mesonephric duct (Wolffian duct), which runs parallel to the fallopian tube [13]. Mesonephric type paraovarian cysts may develop from either the mesonephric duct or tubal remnant [6, 7].

Asymptomatic cases of paraovarian cysts are often discovered by chance during laparotomy. The most symptomatic paraovarian cysts necessitating surgical treatment are those with a diameter over 5 cm [1]. They may sometimes reach a size large enough to cause an increase in abdominal volume and clinical symptoms secondary to displacement of pelvic organs; the patient may also have a feeling of abdominal heaviness [7, 20]. In addition, paraovarian cysts infrequently present symptoms related to complications such as internal haemorrhage from rupture or torsion [1]. The most common symptom of paraovarian cysts is mild or moderate lower abdominal pain. This symptom is similar to the pain associated with a twisted ovarian cyst, although often less severe [1, 7]. Although rare, paraovarian cysts containing neoplasms have been reported [13]; therefore the obstetrician-gynaecologists should have in mind that paraovarian tumors may be malignant [11, 12]. Fortunately, the incidence of malignant paraovarian tumors is low (2-2.8%) [6, 11] and occur mainly during the reproductive years of women [11]. The treatment for paraovarian symptomatic cysts is surgical [20]. In our case, the patient presented with symptoms of a twisted pelvic mass. Additionally, the size of the right paraovarian cyst and the presence of a second cyst on the left ovary aggravated the pulmonary function of the patient. The preoperative computed tomography showed the presence of a huge mass which expanded from the level of the internal genital organs coming in contact with the uterus to the mesocolon. The margins of

the mass were clear and its wall was thin. However, because of the presence of the second cyst in the left ovary the paraovarian cyst was interpreted as bilocular and the presence of an ovarian cystadenoma was suspected.

Large intra-abdominal tumors may produce immobility, breathlessness and an inability to lie supine. Elevation and splinting of the diaphragm lead to tachypnoea [14, 22, 23]. Hypercapnia, hypoxia, mucous plugging and ventilatory failure have been reported because of the patient's inability to breathe deeply, cough and expand her lung bases [24]. Similar to a gravid uterus, such cysts exert pressure on the inferior vena cava, decreasing both venous return and cardiac output [24]. Supine hypotension is common, as cardiac output may drop 50% in the supine position [14, 22, 23]. Venous obstruction also produces oedema of the legs and dilatation of the superficial veins of the abdominal wall [23]. A patient admitted for removal of a large intra-abdominal mass should undergo cardiac and respiratory assessment and function improvement if needed [23]. Preoperative coagulation tests and the availability of four to six units of blood are indicated [24]. Intraoperatively, placement of the surgical table in reverse Trendelenburg will help increase the lung volume [14]. An arterial line is useful for continuous monitoring of blood gases. Acute, life-threatening fluctuations in blood pressure may occur at the time of cyst removal [24]. Both splanchnic shock and acute hypertension, secondary to increased venous return, have been described [25]. Intraoperative haemorrhage is frequently reported due to dilated abdominal varicosities and dense adhesions between the mass and the abdominal wall [24]. In the early postoperative period, when circulation is unstable, central venous pressure and blood volume estimations are helpful in assessing blood and fluid requirements [23]. In regards to surgical management, in patients with large abdominal masses the most appropriate incision and closure technique must fulfill three needs: (a) adequate exposure for removal of the mass without rupture, (b) re-approximation and strengthening of the attenuated, separated layers of the anterior abdominal wall, (c) removal of excess skin [24]. The cysts should be removed intact, whenever possible, in order to avoid intraperitoneal spillage of cyst contents [24]. In this case, both masses were removed intact with good results and the patient showed notable improvement of her respiratory function postoperatively.

In conclusion, we reported a very rare case of an enormous twisted paraovarian cyst in a 74-year-old patient and we presented some basic concepts of management generated during the care of this case. Obstetrician-gynaecologists should bear in mind that paraovarian cysts might reach large sizes even in elderly patients complicating their already aggravated cardiopulmonary function.

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Address reprint requests to:
M. N. VARRAS, M.D., Ph.D.
Obstetrician - Gynaecologist
Consultant in Obstetrics and Gynaecology
Platonos 33 - Politia (Kifisia) 14563
Athens (Greece)