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Giant Paraovarial Cystadenoma in An Adolescent: Case Report and Literature Review

Fedoruc A.
National Scientific-Practical Center of Pediatric Surgery "Natalia Gheorghiu", Moldova

Babuci S.
National Scientific-Practical Center of Pediatric Surgery "Natalia Gheorghiu", Moldova

Gladun S.
National Scientific-Practical Center of Pediatric Surgery "Natalia Gheorghiu", Moldova
State University of Medicine and Pharmacy "Nicolae Testemitanu"

Ambros I.
National Scientific-Practical Center of Pediatric Surgery "Natalia Gheorghiu", Moldova
State University of Medicine and Pharmacy "Nicolae Testemitanu"

Petrovici V.
National Scientific-Practical Center of Pediatric Surgery "Natalia Gheorghiu", Moldova
State University of Medicine and Pharmacy "Nicolae Testemitanu"

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Introduction. Paraovarian cysts are adnexal formations in the mesosalpinx within the broad ligament. They are thought to arise from remnants of the paramesonephric (Müllerian) ducts in approximately 30% of cases, the mesonephric (Wolffian) ducts in about 2% of cases, or from mesothelial tissue in nearly 68% of cases. [1, 8]. These cystic formations can occur in women of all ages, with the peak incidence in the third and fourth decades of life [25]. The reported incidence of paraovarian cysts in children and adolescents is approximately 4–7.3%. [23, 25, 34].

The average size of paraovarian cysts is approximately 7.5 cm, with 95% measuring less than 2.0 cm in diameter. There is no consensus on the definition of a giant cyst [7, 19]. Only 12.96% of these cysts exceed 15 cm, which some authors consider as giant [8, 28], while others propose a diameter of 20.0 cm as a more appropriate threshold [36]. In adolescents, the size of giant cysts reported in the literature has ranged from 17 to 40 cm, making the differential diagnosis challenging. Potential differentials include benign or malignant ovarian tumors, tubal formations, tubo-ovarian abscesses, and non-gynecological abdominal cystic lesions [92]. Few cases of giant paraovarian cysts in the pediatric population have been published, all managed with different surgical approaches [26]. In this context, we present a case of an adolescent girl with a giant paraovarian cyst, which posed diagnostic challenges on imaging.

Case presentation. Patient A., a 16-year-old girl, presented to the hospital with complaints of intermittent abdominal pain that began 5 days prior, accompanied by abdominal distension. On physical examination, a large, tender, and moderately mobile

mass was palpated in the lower abdomen, extending into the mesogastric region.

The patient's medical history revealed no history of sexual intercourse, no use of hormonal medications, and no prior surgical interventions.

Initial investigations included abdominal ultrasonography, laboratory tests, and magnetic resonance imaging (MRI). Ultrasonography revealed an intra-abdominal cystic formation of unknown etiology, measuring approximately 13 × 10 cm. Results of complete blood count, biochemical analyses, and urinalysis were within normal reference ranges. Serum tumor marker testing showed normal α-fetoprotein levels at 6.20 ng/mL (reference range 0–20 ng/mL), whereas hCG was elevated at 16.22 mIU/mL (reference range 0–10 mIU/mL).

MRI revealed a unilocular cystic formation with homogeneous fluid content similar to cerebrospinal fluid, without septations, located in the anterior pelvic compartment and extending into the abdominal cavity up to the L3 level. The cyst was elongated in the transverse plane, well-defined, and measured 18 cm (vertical) × 6.6 cm (anteroposterior) × 20 cm (transverse). The lesion caused a marked mass effect on the uterine adnexa, uterus, bladder, and intestinal loops, without evidence of intramural nodules, fat, or calcified components (Fig. 1).

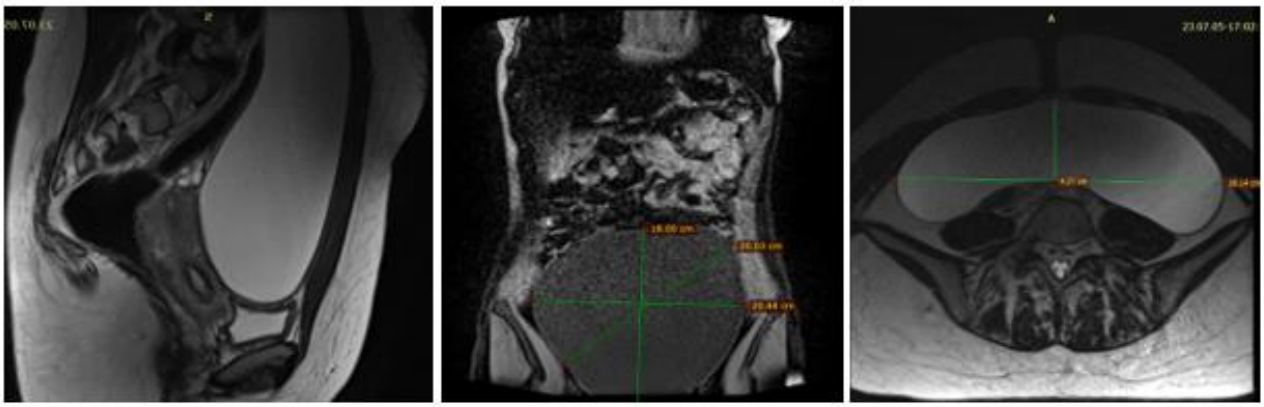

Fig. 1. Patient A. aged 17. Preoperative MRI: cystic, single-chamber, purely fluid formation with signs of pronounced mass effect on uterine adnexa, uterus, bladder, bowel loops (explanation in text).

Considering the large size of the cystic formation and the uncertain nature of the tumor (benign or malignant), an inferior midline laparotomy was performed instead of laparoscopy. The choice of an open abdominal approach was also supported by the imaging findings, with a mesenteric cyst included in the differential diagnosis. Upon opening the abdominal cavity, a large cystic formation was identified, attached medially to the ampulla of the fallopian tube. The cyst was punctured, releasing approximately 1.5 liters of yellowish serous fluid. After exteriorization, the lesion was confirmed to be paraovarian on the right side, without adhesions to adjacent organs. The right ovary was intact, while the left ovary showed polycystic changes. The cyst was removed en bloc within its own capsule (Fig. 2). Subsequently, the broad ligament on the right side was reconstructed and peritonized. The procedure concluded with restoration of normal anatomical planes.

Fig. 2. Intraoperative aspects of mobilization of the cystic formation (A, B). C - macroscopic aspect of the removed cyst.

The postoperative course was uneventful, and the patient was discharged in satisfactory condition on the seventh postoperative day. At one-month follow-up, she reported no complaints, and the surgical wound had completely healed.

Macroscopic examination of the specimen revealed a flaccid cystic formation with a “pear” shape and a pearly surface, featuring non-transparent areas with prominent vascular markings. On dissection, the cyst contained transparent serous fluid. The internal surface was pearlescent like the external surface, mostly opaque, occasionally semi-transparent, with prominent vascular markings. A circular fold of firm, elastic consistency extended across the entire internal surface, corresponding to the boundary between the disproportionate volumes of the cyst (Fig. 3).

Fig. 3. Macroscopic aspects of the removed paratubal cyst: A - pear-shaped cystic formation, externally pearly, flaccid appearance after partial removal of serous fluid; B - dissected cyst: transparent serous content in the cavity, at the boundary between the cyst volumes a fold (black arrow); C - fold on the internal surface of the cyst having the same thickness as the cyst wall

Histological examination revealed a fibrillar connective wall containing spindle-shaped cellular elements of mild to moderate density. The internal surface was frequently lined by a cuboidal or columnar pseudo-stratified, sometimes multi-layered ciliated epithelium. In some areas, Hobnail cells (cells with clear cytoplasm), characteristic of tubular epithelium, were observed (Fig. 4A, B). The fold region exhibited a structure similar to the cyst wall, with an epithelial lining analogous to that found in other areas of the cyst, showing a pseudo- or multi-layered pattern. Adjacent to the fold, a small plateau with stromal-epithelial papillary structures exhibiting minimal proliferation was noted (Fig. 4C, D).

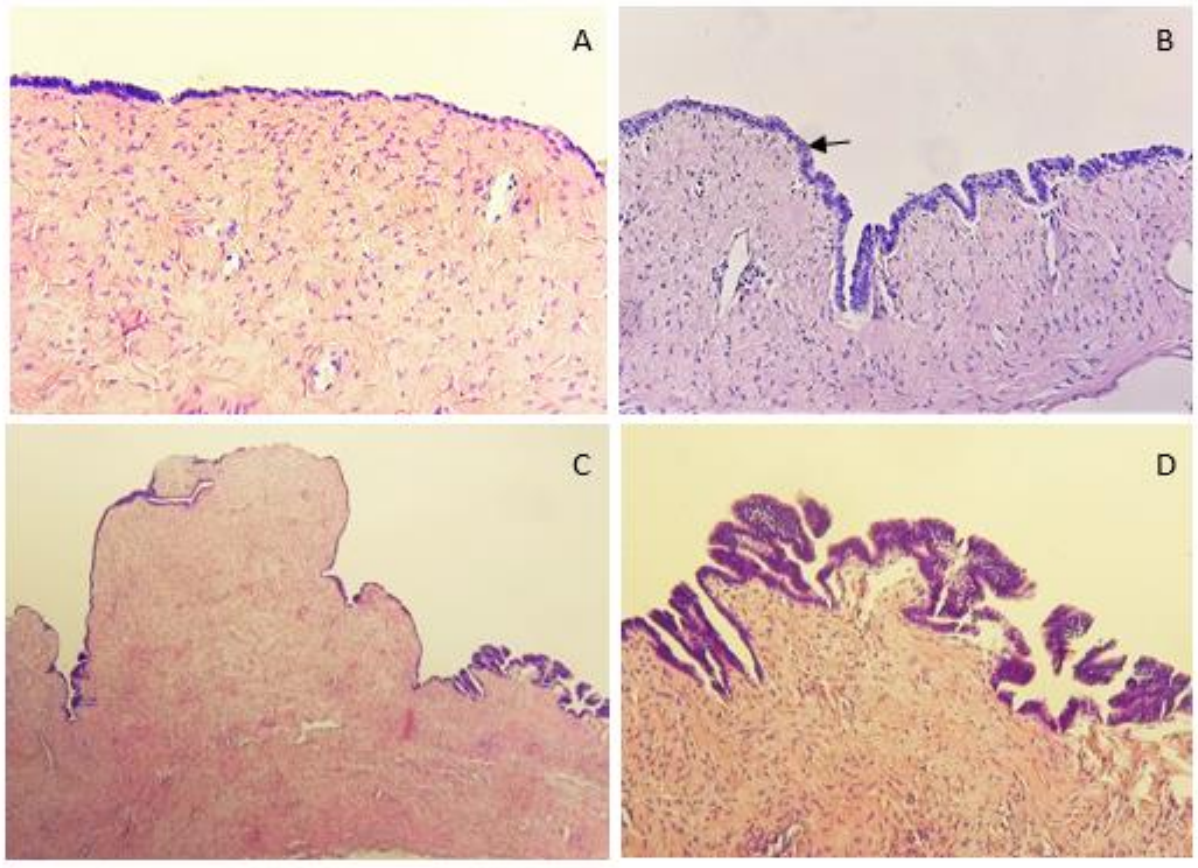

Fig.4. Histopathological aspects of the cyst wall. A - mixed pseudo-stratified (a) and cuboidal (b) surface epithelium. Colour. HE, 200x; B - epithelium with features of tubular origin with the presence of Hobnaila cells (arrow). Color. HE, 200x; D - small papillary structure of the fold in the apical area and an adjacent small plateau of papillary stromal-epithelial papillary structures. Color. HE, 200x; D - small waist plateau with fibro-epithelial structures lined with ciliated pseudostratified columnar epithelium. Color. HE, 200x;

Of note, no mitotic activity, dysplasia, or atypia of the cystic epithelium was observed. Portions of the fallopian tube showed stasis congestion, stromal edema, and, in some areas, lymphocytic infiltration and sclerotic changes. Taken together, these histological findings confirm the diagnosis of a benign paraovarian cyst, represented by a focal micropapillary serous cystadenoma.

Discussions. A paratubal or paraovarian cyst is a fetal-origin lesion, first described as a distinct pathological entity by Kariminejad M.H. and Scully R.E. in 1973 [16]. The terms “paratubal” and “paraovarian” are used according to the proximity of the cyst to the fallopian tube or ovary, respectively. [35].

According to some authors, only six cases of giant paraovarian cysts, ranging in size from 17 to 20 cm in adolescent girls, were reported by 2019, and laparoscopic surgery was performed in only one of these cases [18].

Paraovarian cysts are predominantly asymptomatic, which is why only 30–44% are identified preoperatively [12], and in 15.7% of cases they are discovered incidentally during diagnostic laparoscopy [6]. During the course of the condition, patients may experience lower abdominal heaviness, mild pain, or urinary symptoms, which in some cases may be accompanied by fever [15].

Occasionally, clinical complications may arise due to progressive enlargement of the cystic formation, including hemorrhage, torsion, cyst rupture with hemoperitoneum, torsion of the tumor or ipsilateral fallopian tube, or malignant transformation [17, 24, 33, 34].

Paratubal cysts may be simple or of neoplastic origin [14]. Simple paraovarian cysts arise from embryonic

remnants of the urogenital system (mesonephric and paramesonephric ducts) or from invagination of the serosa of the fallopian tubes, forming mesothelial cysts. Neoplastic paraovarian cysts develop either from neoplastic transformation of a simple paraovarian cyst or from the adjacent ovary [10]. The majority of paratubal cysts are benign, with malignancy reported in approximately 2–2.9% of cases; the main secondary neoplasms are cystadenocarcinoma and papillary carcinoma [31, 37]. However, some studies have reported a higher incidence of neoplastic paraovarian cysts, around 25%, usually diagnosed in adults [22, 29]. Histological examinations have also revealed, in addition to simple paraovarian cysts, cases of cystadenofibroma, cystadenoma, adenomatoid tumors of mesothelial origin, and borderline papillary serous tumors [14, 27]. Cases of borderline tumors arising in paratubal cysts in adolescent girls have been reported [20]. For differential diagnostic purposes with epithelial tumors, it is recommended to assess tumor markers such as cancer antigens (CA-125, CA 15-3, CA 19-9), inhibin B, beta-human chorionic gonadotropin (β-HCG), carcinoembryonic antigen (CEA), lactate dehydrogenase (LDH), alpha-fetoprotein (AFP), estradiol, and others [3, 21]. According to some studies, these markers are elevated in only 54% of malignant adnexal neoplasms and in about 6.5% of benign lesions [30].

Preoperative differential diagnosis between ovarian and paratubal cysts remains challenging, as pathognomonic imaging features specific to paraovarian cysts are incompletely described [35]. According to some studies, a definitive preoperative diagnosis of a paratubal cyst by ultrasound is achieved in only 25–44% of cases, whereas color Doppler ultrasound demonstrates a specificity of 99% but a sensitivity of only 14% [17, 25]. Some authors consider ultrasonography and magnetic resonance imaging (MRI) to be more useful than computed tomography (CT) [1]. According to a consensus statement, regardless of patient age, cysts larger than 7 cm warrant further evaluation with MRI [14].

The differential diagnosis of paraovarian cysts includes intestinal duplication, internal hernias, intestinal lymphangioma, and peritoneal inclusion cysts [32].

There is no consensus regarding the surgical management of paraovarian cysts [36]. No specific criteria have been established for choosing between laparoscopy and laparotomy; surgical techniques are adapted according to cyst size, risk of intraoperative rupture, patient age, concomitant pathologies, and other factors [3, 4, 11, 38]. Treatment options for paratubal cysts include cystectomy, cystectomy with salpingectomy, or, in complicated cases, adnexectomy [2, 3, 5, 13]. Aspiration of the cystic fluid must be performed with great precision to avoid intraperitoneal spillage, which could result in tumor seeding in the peritoneal cavity in cases of malignant cystic lesions [11].

Thus, this clinical case report highlights the rarity of a large benign paratubal serous cystadenoma diagnosed in adolescent girls, as well as the challenges associated with preoperative diagnosis. In the present case, laparotomy was preferred over laparoscopy due to the large size of the cyst and the risk of intraperitoneal dissemination of its contents. The presence of papilliform tissue on the inner surface of the cyst should be carefully considered, as some neoplasms, including borderline tumors, cystadenocarcinoma, and papillary carcinoma, may arise from this tissue.

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