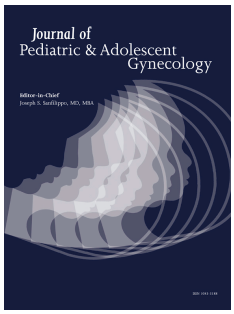

Giant paratubal serous cystadenoma in an adolescent female: Case report and literature review

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Running title:

Giant paratubal cystadenoma in an adolescent female

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Abstract

Background: Paraovarian/paratubal cysts constitute 5-20% of all adnexal lesions and typically originate from the paramesonephric or Müllerian duct. The primary epithelial tumors arising from paraovarian cysts account for 25% of the cases, but giant cystadenomas of paraovarian origin are extremely uncommon during childhood and adolescence with very few cases reported in the literature.

Case: We present the case of a 15-year-old female that presented with a bulky mass in the abdomen and pelvis. An initial clinical and radiological examination indicated an ovarian cyst measuring ~25x20 cm. However, explorative laparotomy revealed a giant paratubal cyst that was successfully treated with complete excision using fertility-sparing surgery. Histopathological examination was consistent with a serous cystadenoma. The postoperative course was uneventful and the girl was discharged on the seventh postoperative day. At the follow-up of six months, the patient was doing well.

Summary and Conclusion: Due to their rarity and enormous size, the proper diagnosis and adequate management of giant paratubal cystadenomas are challenging. A complete excision of cystadenoma with preservation of adnexa represents a desirable treatment modality in adolescent females and should be attempted whenever possible.

Key words: Paratubal cysts, serous cystadenomas, adolescence, diagnosis, surgery

Introduction

Paraovarian masses are a relatively common finding, accounting for 5–20% of all adnexal lesions [1-3].

They may be either non-neoplastic simple cysts or cysts of neoplastic origin [4]. The simple paraovarian cysts arise from the broad ligament between the Fallopian tube and the ovary and their origin may be mesothelial, mesonephric, or, more commonly, paramesonephric (Müllerian) [5]. It was previously believed that the most neoplastic paraovarian cysts originate from a neoplastic transformation of paraovarian simple cysts or from the adjacent ovary[2], but more recently cystadenomas and cystadenofibromas are thought to develop *de novo* from a single cell into a cystic lesion.

The neoplastic paraovarian cysts originate from a paraovarian simple cyst or from the adjacent ovary and are generally benign serous cysts (cystadenomas) similar to benign ovarian tumors (i.e., cystadenomas or cystadenofibromas)[2]. Paraovarian tumors of borderline malignancy or malignant paraovarian tumors are very rare [3,4,6]. Serous cystadenomas (SCAs) are uncommon neoplasms among pediatric and adolescent females (~3%) and the available data come from case reports and small case series[2]. These lesions are usually large at presentation causing clinical symptoms due to a compressive effect on adjacent organs [3]. The symptoms can also occur due to complications caused by torsion and internal hemorrhage from rupture in the form of an acute onset of abdominal pain or irritation of the peritoneum and less frequently as a circulatory collapse and hemorrhagic shock [7].

Herein we present the case of a 15-year-old adolescent female with a giant left-sided paratubal SCA that presented with a bulky mass in the abdomen and pelvis. The mass was successfully treated with a complete paratubal cystectomy using a fertility-sparing procedure.

Case

A 15-year-old postmenarchal female was admitted for evaluation of a 3-month history of a gradual asymmetric abdominal enlargement, mainly in the hypogastric region, followed by intermittent nonspecific abdominal pain, and constipation for up to 3 days. Her medical history was uneventful and she had reported regular menstrual cycles. She had achieved menarche at the age 12. At admission, the patient was hemodynamically stable as her blood pressure was 110/70 mm Hg and pulse rate 78 beats/min. A physical examination was remarkable for a smooth, firm, and painless abdominal mass, extending from the pubis to 2-3 cm above the umbilicus (Figure 1C). Her secondary sexual characteristics corresponded to her age according to the Tanner scale. Imaging modalities including abdomen ultrasound (US) and magnetic resonance imaging (MRI) revealed a huge abdominal-pelvic cystic lesion arising from the left adnexa. The cyst measured 20.5 × 8.4 cm on the cross-section and about 25 cm in length (Figure 1A-B). It had a thin wall and contours without papillary proliferations, all of the features suggestive of its benign nature (Figure 1A-B). The left ovary was not separately visualized while the right ovary was normal. No free fluid in the abdomen and pelvis was observed. The values of the serum tumor markers were within normal range[8]: Lactate dehydrogenase (LDH): 180 U/L, Alpha-Fetoprotein (AFP): 1 ng/mL, Cancer antigen (CA) 125: 5.2 U/mL, and β-human chorionic gonadotropin (beta-hCG): 2.4 mlU/ml.

At open surgery via a 5 cm low transverse Pfannenstiel incision, the uterus and both ovaries were normal in appearance. A huge (~25 cm) paratubal cyst arising from the left mesosalpinx and occupying the entire pelvis and lower abdomen was found (Figure 2A-B). The cyst wall was intact and adhesion-free without any solid components or external excrescences. After covering the lesion with sterilized adhesive surgical sheet to prevent the leakage, the cyst was carefully punctured using a suction irrigation apparatus and a total of 3.8

L of serous fluid were aspirated without any spillage from the cyst, allowing the decompressed cyst and adnexa to be externalized. During the surgery, we had a marked dilemma on how to deal with the highly elongated left Fallopian tube. Although we were worried about possible complications related to an unattached tube to the left ovary, due to the inability to adequately attach the ovary tube, we decided to leave the elongated tube free in the pelvis with close postoperative follow-up. Therefore, the patient underwent left paratubal cystectomy using a fertility-sparing procedure with a complete preservation of both ovaries and fallopian tubes (Figure 2A-B).

The specimen was submitted to the histopathology that confirmed a serous cystadenoma arising in a paratubal cyst (Figure 2C-D). The postoperative course was uneventful and the girl was discharged on the seventh postoperative day. At the follow-up of six months, the patient was doing well.

All the procedures followed were in accordance with the ethical standards of the Helsinki Declaration of 1975, as revised in 1983. The patient also gave consent to publish the data presented in the case study. The local institutional review board (IRB) has the policy not to review the case studies.

Summary and Conclusion:

Only a few cases of giant paratubal SCAs have been reported in the literature so far and most of those cases are related to adult women [9,10]. In contrast to adult women, epithelial tumors are much less common in children and adolescents. The tumors are usually serous or mucinous and classified as benign (70%), borderline (5-10%), or malignant tumors (20-25%) [11,12]. Histological types of the benign paraovarian tumor include serous cystadenoma, papillary serous cystadenoma, serous cystadenofibroma, mucinous cystadenoma, and endometrioid cystadenoma. Benign tumors are usually unilateral, cystic, mobile, and smooth [11]. Most of these tumors produce mild, non-specific symptoms including abdominal distension, intermittent abdominal pain or discomfort and lower abdominal pressure sensation, and in some cases, symptoms affecting the gastrointestinal or urinary tract [3].

Despite the advances in preoperative diagnostics, an accurate diagnosis of adnexal masses is still difficult and challenging. In addition, radiological approach to the adnexal masses, primarily paratubal cysts, is still not uniformly reported[13]. However, the size, persistence, and separability from the adjacent ovaries are the most helpful clues for identification of nonphysiological paratubal cysts[13].

Unlike ovarian cysts in premenopausal women, which are mostly functional and regress without treatment or less frequently treated with cyst puncture, combined oral contraceptive pill, hormonal replacement therapy and surgery, the treatment of choice of SCAs is a surgical excision owing to the risks of spontaneous rupture, torsion and/or malignancy. Paratubal cystectomy is technically easy and is feasible in almost all cases. We demonstrated that a fertility-sparing procedure could also be performed successfully with large SCAs when the diagnostic findings suggest their benign nature. Others have suggested this approach as well [3,14].

In conclusion, giant paratubal SCAs in adolescent females are extremely rare, but have an excellent prognosis as confirmed in our case. A fertility-sparing surgery should be a preferable treatment method and attempted whenever possible.

Disclosures

The authors have no conflicts of interest to disclose.

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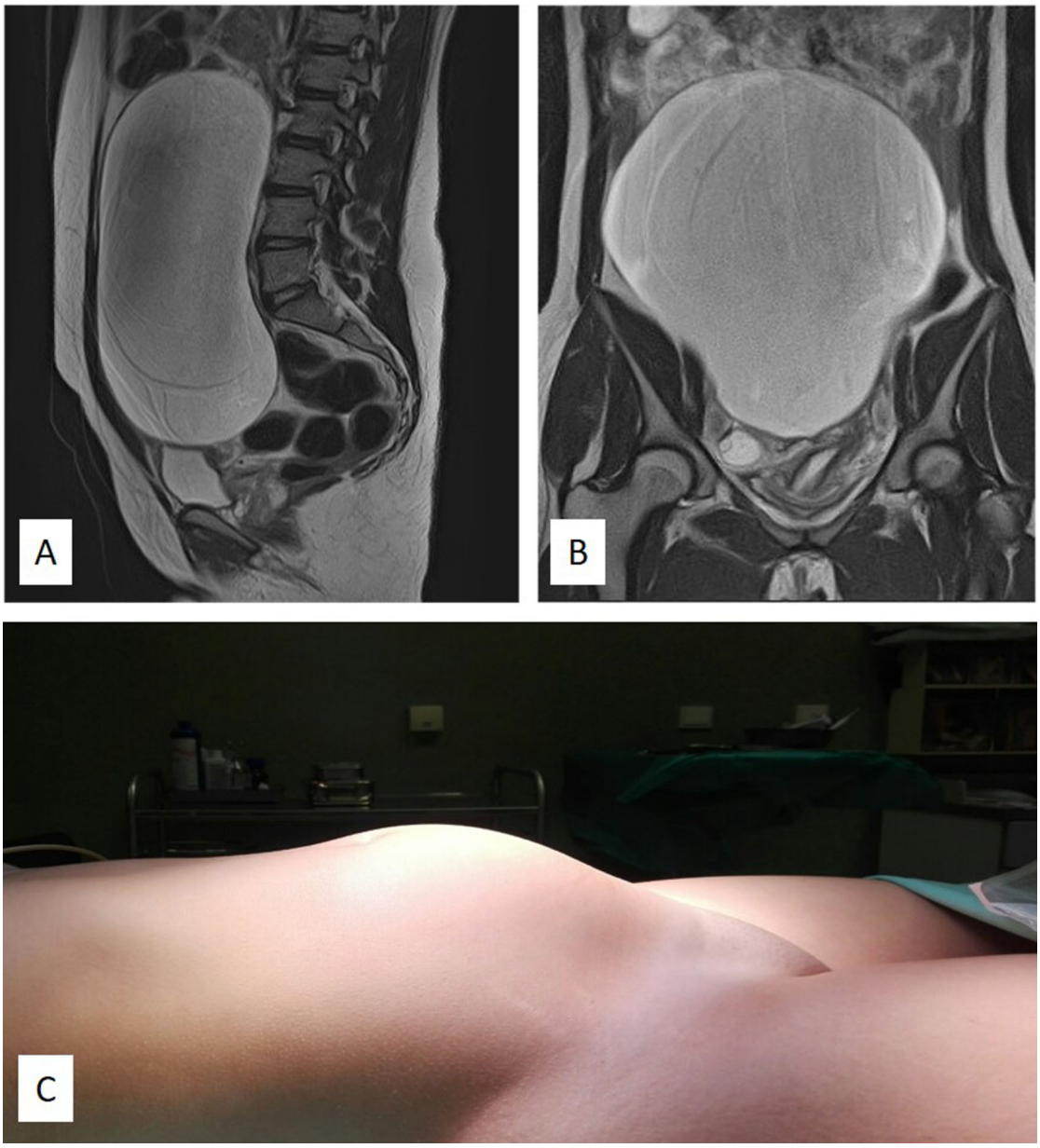
Figures

Figure 1A-C: Contrast enhanced coronal and sagittal MRI view of a giant unilocular paratubal cyst (A-B). Preoperative view of distended abdomen due to left-side paratubal cyst (C).

Figure 2A-D: Intraoperative view of the giant paratubal cyst capsule after aspiration of 3,800 mL of clear, serous fluid (A); Intraoperative view of the left ovary and elongated left fallopian tube after paratubal cystadenoma removal (B); Histopathological examination revealed a cyst with dense fibrous stroma and simple papillary projections on its surface (C) (Hematoxylin and Eosin stain, 4x magnification). The papillary projections were lined by columnar and cuboidal epithelial cells resembling normal tubal/ovarian surface epithelium (D) (Hematoxylin and Eosin stain, 10x magnification).


A
B
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