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Surgical management of a giant paratubal cyst: a case report and review of the literature



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Abstract

Background Surgical management of giant paratubal cysts can be challenging in the pediatric and adolescent population. Larger adnexal masses can heighten suspicion for malignancy, placing the ovary at risk of unwarranted removal, and larger masses sometimes necessitate larger surgical incisions which can increase postoperative pain and delay return to school and regular activities. We present the evaluation and management of a giant paratubal cyst containing 16 l of fluid in an adolescent, using a mini-laparotomy approach.

Case presentation A 16-year-old patient presented with 1-month history of worsening nausea and increasing abdominal girth. Abdominal imaging revealed a 42-cm adnexal mass. A modified drainage technique with a mini-laparotomy incision was utilized, thereby preventing tumor spill while minimizing size of the surgical incision. Postoperative recovery was complicated by prolonged nausea, but the patient was discharged home 2 days after surgery and returned to school the following week.

Conclusion This case illustrates that giant adnexal masses can be managed using a mini-laparotomy approach, thereby expediting return to regular activities.

Keywords Giant paratubal cyst, Serous cystadenoma, Minimally invasive technique

Background

Paratubal cysts are typically benign adnexal masses, frequently less than 3 cm in size, and originate from paramesonephric or mesonephric ducts [1]. The Mullerian (paramesonephric) and Wolffian (mesonephric) ducts coexist early in embryologic development, and during the involution process, remnants of these structures invaginate and can become cystically dilated leading to the formation of paratubal cysts. Paratubal cysts have been reported to comprise approximately 5% of all adnexal masses, with a higher incidence of up to 7.3% in the

pediatric and adolescent population [1, 2]. The mean size of the paratubal cysts in the adolescent cohort was 6.13 cm, with a range of 0.5 to 33 cm [2]. These cysts are commonly found in children and adolescents during surgery for adnexal torsion. Based on their embryological origin, it is theorized that these cysts are less likely to recur, but there is little data in the published literature to guide management [2].

While small paratubal cysts are relatively common, giant paratubal cysts of > 25 cm are rare, with only a few cases noted in the literature [3–5]. As adnexal masses increase in size, so too does the risk of malignancy. While the majority of paratubal cysts are benign, the literature has reported the incidence of malignancy among those diagnosed with paratubal or paraovarian cysts to be as high as 2–3%. Most of these cases were paraovarian rather than paratubal, associated with intrapapillary excrescences, and had other findings suggestive of malignancy [6].

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With increasing frequency, large adnexal masses with a high likelihood of benign final pathology are managed with minimally invasive techniques including laparoscopic-assisted drainage or drainage through a mini-laparotomy incision, in order to optimize surgical outcomes and return to baseline function [4, 7–9].

We present a case of a 42-cm paratubal cyst, containing 16 l of serous fluid. To our knowledge, this is one of the largest paratubal cysts reported in the pediatric and adolescent literature managed with a less invasive approach.

Case presentation

A 16-year-old individual presented to primary care with worsening nausea and increasing abdominal girth for 1 month. Medical history was significant for gastroesophageal reflux disease (GERD), generalized anxiety, and recurrent periodic urticaria. Gynecologic history was notable for menarche at age 12, with cyclic menses every 28–34 days, lasting 5–7 days. The patient reported no family history of malignancy and no prior surgeries. Regarding medications, the patient reported taking overthe-counter famotidine daily for GERD, ondansetron as needed for nausea related to dyspepsia, and lorazepam as needed for generalized anxiety. On physical examination, vital signs were within normal limits. Abdominal examination was notable for a distended and firm but mobile, non-tender cystic mass, with multiple striae across the abdomen (Fig. 1). In-office testing returned a negative urine pregnancy test. Abdominal ultrasound reported a complex fluid collection involving the pelvis and most of the abdomen that was noted to be too large to measure by ultrasound and therefore concerning for underlying ovarian tumor (Fig. 2A). The patient was also noted to have right urinary tract dilation and hydroureter. The primary care physician recommended transfer to the emergency department (ED) for further evaluation.

In the ED, the patient's vital signs and abdominal exam remained stable. Computed tomography (CT) scan revealed an extremely large (41.6 \times 28.2 \times 21.2 cm) hypodense lesion extending from the pelvis to the upper abdomen, suspicious for an underlying ovarian lesion (Fig. 2B,C). Marked right urinary tract dilation with hydroureter and diffuse cortical thinning was again noted. There was no evidence of lymphadenopathy or omental thickening. Review of ultrasound images with radiology revealed the mass to be entirely cystic in nature, arising from the right adnexa, without evidence of complexity as noted in the original report.

Laboratory evaluation included a complete metabolic panel and a complete blood count, which were within normal limits, including normal renal function. Adolescent gynecology was consulted and recommended obtaining tumor markers given the large size of the



Fig. 1 Preoperative examination of the abdomen

mass and concern for possible underlying malignancy, based on the initial ultrasound report. Tumor markers included cancer antigen-125 (CA-125), inhibin B, betahuman chorionic gonadotropin (β -HCG), lactate dehydrogenase (LDH), alpha-fetoprotein (AFP), and estradiol, which were all within normal limits. Based on the normal tumor markers and markedly enlarged but simple nature of the cyst, the most likely diagnosis was deemed to be a paratubal or paraovarian cyst. Other etiologies that were considered included serous cystadenoma, mucinous cystadenoma, borderline tumor, peritoneal inclusion cyst, and less likely germ cell tumor, mucinous or serous cystadenocarcinoma.

Intraoperatively, given the large but simple nature of the mass, decision was made to proceed with a minilaparotomy incision with careful decompression of the mass to prevent tumor spill. A 6-cm vertical incision was utilized to allow for further extension of the incision as needed, should malignant findings be encountered. Once the peritoneal cavity was entered, the cyst wall was identified, and a sterile ultrasound probe cover attached to the surface of the cyst using 2-octyl cyanoacrylate (Dermabond[™]) in order to prevent spill of tumor fluid. A Veress needle was then attached to a Neptune suction machine to allow for rapid mass decompression and was inserted directly into the mass through the ultrasound bag; Fig. 3 demonstrates the equipment utilized for the cyst drainage. Clear, serous fluid was then drained for a total of 16 l. The mass was then delivered through the incision and confirmed to be a giant paratubal cyst (Fig. 4). The fallopian tube was severely distorted from

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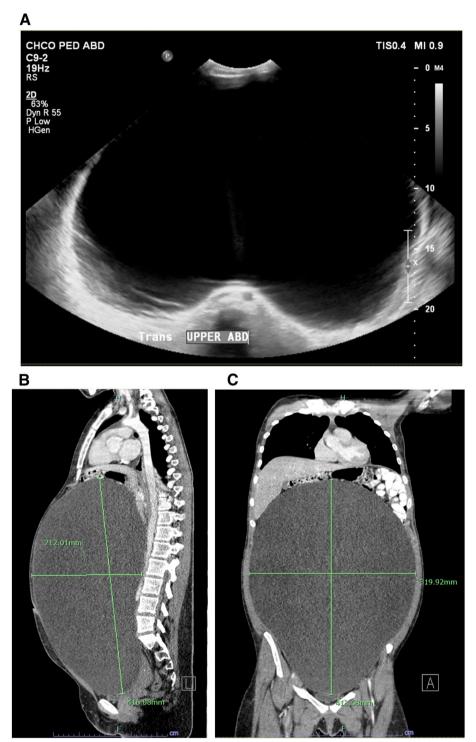


Fig. 2 Preoperative imaging, including abdominal ultrasound A and CT scan B-C, demonstrating a giant adnexal mass

the large mass and deemed unsalvageable, and therefore, a paratubal cystectomy and salpingectomy were performed. Final pathology confirmed a paratubal serous

cystadenoma and an elongated fallopian tube, measuring 15 cm in length.

The patient was discharged on postoperative day 2, requiring a 2-day stay given postoperative nausea and



Fig. 3 Equipment utilized for mass decompression, including Alexis-O abdominal wall retractor, Veress needle, Dermabond adhesive, and sterile ultrasound probe cover



Fig. 4 Decompressed intraoperative paratubal cyst

fatigue. The patient was able to return to school 1 week after surgery and had slow resolution of nausea over the next several weeks, managed with ondansetron and famotidine.

Discussion

First and foremost, it is critical when evaluating large adnexal masses to assess for benign versus malignant features. This is particularly relevant when the size of the mass exceeds 5-10 cm, given the increasing risk of malignancy with larger adnexal masses [1]. The majority of malignant ovarian masses in adolescents are germ cell in origin, and distinguishing physical exam findings is generally lacking in patients with malignant germ cell tumors [1]. However, ultrasonographic features that are more worrisome for malignancy include tumors that are multiloculated, solid, and have thick septae with areas of nodularity, papillary projections, or neovascularization by Doppler flow [1]. Furthermore, computed tomography scanning may show metastatic involvement of the omentum or lymph nodes; however, when disease is limited to the pelvis, sonography is generally superior [1]. In review of our patient case, the only concerning feature was the markedly enlarged size of the mass. Furthermore, upon secondary review of the sonogram, the mass was noted to be simple in nature without loculations, nodularity, or neovascularization. When ultrasound imaging is inconclusive, magnetic resonance imaging can be useful to better delineate soft tissue structures. However, in this particular case, the mass was primarily cystic in nature, and therefore, MRI imaging was deemed unnecessary.

In addition to consideration for additional imaging, when a mass shows some features that are benign and others that suggest malignancy, as was the case with our patient with the large dimensions of the mass, tumor markers can be helpful to guide treatment and surgical approach [1]. Recommended tumor markers include those associated with germ cell tumors, including β -HCG, AFP, LDH, inhibin B, and estradiol, as well as CA-125 for the much less likely epithelial ovarian tumor at this age [1]. In our patient, given the reassuring tumor markers, decision was made to proceed with a mini-laparotomy approach given the likely benign nature of the mass. However, given the very large size and possibility for borderline tumor or early-stage invasive tumor which may be associated with normal tumor markers, the decision was made to proceed with a mini-laparotomy with a vertical incision, which could be easily extended if gross signs of malignancy were discovered intraoperatively.

Despite the enormous size of the paratubal cyst and the remarkable volume of cyst fluid in our patient, we were able to conservatively manage removal of the paratubal cyst using a modified drainage technique. A sterile ultrasound probe cover was chosen to allow for large surface area contact with the cyst. This was selected to allow for rapid decompression of the large volume of cyst fluid and prevention of tumor spill from potential rapid retraction of the cyst wall into the abdominal cavity. This technique is similar to techniques previously described, but the largest masses in these case reports were 20-25 cm in diameter and only drained up to 7000 ml of serous fluid [3-5]. In each of these case reports, surgeons affixed some type of sterilized bag to the cyst wall with a surgical adhesive and drained the cyst through a mini-laparotomy site, measuring 3-6 cm in size across reports [3-5]. Asare et al. described using a sterile ultrasound probe cover affixed to a large cyst wall with Dermabond[™]; however, instead of inserting a Veress needle attached to a large suction device, the team threaded a percutaneous diagnostic peritoneal lavage catheter into the cyst using the Seldinger technique [4]. Zvizdic et al. covered the lesion with a sterilized adhesive surgical sheet through which the cyst was punctured and fluid was aspirated using a suction irrigation device [5]. Shozu et al. attached a polyethylene bag directly onto the cyst wall with cyanoacrylate adhesive, elevated the cyst wall to the abdominal incision, and incised and aspirated the cyst contents [7].

Other groups have managed large cysts with a purely laparoscopic approach. Kilincaslan et al. removed a 20-cm ovarian cyst by percutaneously draining the cyst with a Veress needle before laparoscopically removing the cyst [8]. Another laparoscopic approach used by Coccia et al. involved puncturing the cyst wall with a 5-mm trocar and aspirating the cyst under direct visualization with a suction irrigator while maintaining application of the cyst to the abdominal wall with forceps [9]. More recently, surgeons have described use of the Alexis laparoscopic system®, with demonstration in a case series of six patients with masses up to 30 cm in diameter [10]. After making an initial abdominal incision ranging in size from 2.5 to 4 cm, a small Alexis® retractor was inserted into the abdomen, the cyst was punctured with the trocar, liquid evacuated after trocar insertion into the mass, and the puncture site closed with an Endoloop[®] ligature. The abdomen was then insufflated with gas and the cystectomy completed laparoscopically.

These varied laparoscopic methods allow for minimally invasive removal of large adnexal pathology and shorter postoperative recovery. However, the laparoscopic techniques create the potential for risk of tumor spill around the puncture sites, which can result in upstaging of the tumor and worsening prognosis if malignancy is confirmed. Furthermore, laparoscopic

entry requires abdominal space in order to safely place the laparoscopic trocars and instruments, which was not an option for this patient given the markedly enlarged adnexal mass, extending up to the xiphoid (Fig. 2B,C).

Finally, it should be noted that care should always be taken in the adolescent and young adult patient to preserve future fertility, which includes preservation of the ovary and fallopian tube whenever possible [3, 4]. In our case, although the ipsilateral ovary was spared, the fallopian tube was markedly distorted by the giant paratubal cyst and was not salvageable after removal of the paratubal cyst. Decision was therefore made to proceed with unilateral salpingectomy, in addition to paratubal cystectomy.

Conclusion

This case highlights two unique aspects of managing giant adnexal pathology — preoperative imaging and tumor markers to help delineate risk of benign versus malignant nature of the mass and the importance of a minimally invasive approach for adnexal pathology that avoids tumor spill and optimizes surgical recovery.

Abbreviations

ED Emergency department
CT Computed tomography
MRI Magnetic resonance imaging

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Not applicable.

Disclaimer

Our casereport was approved by the patient of interest, and a consent form was signedby the patient agreeing to publish the report.

Authors' contributions

All authors contributed to the conception and design of this case report. SA prepared the manuscript. VIA and PSH contributed to revisions and final manuscript preparation. The authors read and approved the final manuscript.

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Declarations

Ethics approval and patient consent to participate

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Consent for publication

The patient and family agreed in writing to the use of information pertaining to this case, including all images and photographs, for educational purposes.

Competing interests

The authors declare that they have no competing interests.

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