Recurrent Bilateral Mucinous Cystadenoma: Laparoscopic Ovarian Cystectomy with Review of Literature

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Abstract

The second most common epithelial tumor of the ovary is the mucinous tumors, and it constitutes about 8–10% of all ovarian tumors. The recurrence of mucinous cystadenoma is very rare after complete excision. Few cases have been reported. The case presented had initial surgery for adnexal mass diagnosed as mucinous tumor, performed by laparotomy and was followed up. After recurrence, the patient underwent laparoscopic evaluation and bilateral ovarian cystectomy was performed as a fertility preservation for the patient young age. The histopathological diagnosis was mucinous cystadenoma, the same as the initial one. Management in young patients is challenging, especially in the case of recurrence. Follow-up of these patients is very important and transvaginal ultrasound seems to be currently the most effective diagnostic tool for the follow-up of young patients treated with cystectomy for benign mucinous cystadenomas. Total hysterectomy and bilateral salpingo-oophorectomy is recommended after completing family size or reaching age of 35 for fear of progression or incompliance.

Keywords Recurrent · Mucinous · Cystadenoma · Laparoscopy · Cystectomy

Introduction

Benign tumors of the ovaries are of epithelial origin in 50% of cases. Mucinous tumors are the second most common tumor of the epithelial ones, about 8–10% of the ovarian tumors. They may reach massive dimensions, and the size is not included as a criterion for malignancy; in 77–87% of cases, it is benign. They tend to be cystic, and 76% are multi-locular [1].

Conservative surgery as ovarian cystectomy may be preferred in patients who desire to retain their fertility and young age ones. However, when facing a huge mass, saving the ovarian tissue may be difficult. If cystectomy is not completed thoroughly, recurrences may occur. Only few cases have been reported regarding recurrent benign mucinous cystadenoma. The presented case had initial conservative surgery and was followed up for recurrence. This report discuss this patient

who underwent a laparoscopic conservative surgery after recurrence.

Case Report

The patient was a 24-year-old, gravida 3, parity 2, abortion 1 woman who underwent follow-up at another medical center. The patient underwent right ovarian cystectomy through laparotomy (since 2 years) for 12 cm ovarian cyst. Histopathology revealed mucinous cystadenoma.

She was admitted to our hospital on August 2015 with abdominal pain and distension. She was on combined oral contraceptives since 2 years and has regular menstruation. She was married since 6 years. The patient was vitally stable. Examination revealed pelviabdominal mass, mainly on the right side and separable of the uterus (which was 24 weeks of gestation).

Transabdominal and transvaginal ultrasonography revealed right cystic mass with septations about 17 cm with homogenous contents. The patient's tumor markers CA-125 and CA 19.9 were within the normal ranges while CEA was not available and the patient refused to do it for financial reasons. CT revealed (Fig. 1) a large pelviabdominal cyst about 17 × 16.5 × 9 cm with homogenous contents suggestive of right ovarian cyst, with no ascites.

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Except for the abovementioned findings, the CT was otherwise normal.

The patient underwent laparoscopic management under general anesthesia using four trocar techniques where the primary trocar was inserted through the umbilicus. A large multi-locular cyst originating from the right ovary about 17 cm with another one 7 cm cyst from the left ovary was detected. There was no free fluid in the abdomen with no deposits seen all through the abdomen and pelvis. The intra-abdominal space was washed with an isotonic solution and the aspirate was sent for cytological analysis. It revealed no malignant suspicious cells. The right fallopian tube and ovary were adherent to the peritoneal surface of the lateral pelvic wall. The adhesions were dissected. The omentum and the liver looked normal. Bearing in mind that the patient had desire for fertility, being so young in age, and that this was most commonly a recurrent cystadenoma (history of cystectomy, multi-locular, bilateral, large size, and cyst on top of COC), bilateral ovarian cystectomy was performed laparoscopically. Lateral trocars were inserted directly in the cysts that were aspirated for decompression and sent for histopathology. Dissection of the cyst wall from the ovary was done. Extraction of the cyst wall through lateral 10 mm trocar was done and was sent for histopathology assessment. (Fig. 2) The final pathological result revealed bilateral ovarian mucinous cystadenoma. The patient was discharged from the hospital the next day without any complication.

Discussion

There is limited data regarding recurrent mucinous cystadenomas. Few cases have been reported in the literature. The first case was presented by Olesen and Eisum [2] in 2001. The authors reported a premenarchial girl who underwent laparotomic cystectomy due to a recurrent mucinous cystadenoma [2]. Gotoh et al. [3] published the second case in 2004 who had received ovulation induction therapy after a 5-year period of primary infertility. The patient underwent hysterectomy and bilateral salpingo-oophorectomy after numerous cystectomies failed [3]. The third was presented by Baksu et al. [4] in 2006 from a 20-year-old patient who underwent a left-sided salpingo-oophorectomy and right-sided cystectomy. The patient had a right-sided cystectomy after the first recurrence and finally a total abdominal hysterectomy and right-sided salpingo-oophorectomy upon the second recurrence [4], and a recent report by Mittal et al. [5] described a 25-year-old nulliparous female with a huge benign mucinous cystadenoma managed by laparoscopic cystectomy, followed by recurrence within 2 months. Left-sided salpingo-oophorectomy was performed on a repeat laparoscopy due to suspicion of malignancy on ultrasound. Pathology revealed a benign cyst.

Baksu et al. in 2006 reported a 20-year-old patient that had three laparotomies resulting in the removal of one ovary with a mucinous cystadenoma and two cystectomies for the same pathology, but ultimately leading to hysterectomy and salpingo-

Fig. 1 CT revealing large multi-locular pelviabdominal cyst

Fig. 2 Laparoscopic view revealing bilateral ovarian multi-locular ovarian cyst (a, b), postcystectomy view (c), and cyst wall specimen (d)

oophorectomy [4]. Turkyilmaz et al. in 2009 reported a case that had her initial surgery performed by gynecologic oncology team by laparotomy and was followed up by the same group. After recurrence at the same ovary, the patient underwent laparoscopic evaluation and unilateral salpingo-oophorectomy was performed [6].

Surgical approach for ovarian cysts depends on the patient's age, parity, and the size and structure of the cyst [7]. Similar to the case of Mittal et al. [5], we performed laparoscopic surgery for our patient. As a result, the patient suffered minimal pain and was discharged from the hospital on the next postoperative day without any complication. Mucinous tumors may contain foci of low differentiated epithelium [1]. As they may reach huge sizes, this is a real problem for both frozen and definitive pathological analysis. As it has been recently outlined by Taskiran et al. [8], the rate of misdiagnosis during frozen section is around 17% for mucinous tumors, whereas it is around 5% for the remaining epithelial tumors ($P=0.003$). Therefore, multiple slices must be obtained and evaluated when a mucinous tumor is confronted, to discern evident anaplastic changes and exclude borderline tumors. Because of microscopic similarity between both benign and malign cystadenomas, it is often thought that malignant tumors are derived from benign tumors and the benign ones progress to the malignant ones later with conservative surgeries [9].

An important question related to the recurrent mucinous cystadenomas is whether or not these cases underwent sufficient surgical excision in the first surgical procedure. In our case, the initial procedure was not carried out by our team. Incomplete resection may be one of the hypotheses for recurrence. Although they are classified as benign, there is a risk of recurrence, especially in huge multi-loculated

mucinous cysts. Therefore, close follow-up is required for early detection of recurrences and for rendering conservative and laparoscopic management possible before the cyst reaches huge dimensions.

It is important to keep in mind the possibility of recurrence even of benign cysts. This is especially true for mucinous tumors as they are common, usually benign, and most of the time multi-locular. Because the management of young patients is challenging, the most important issue is probably the follow-up of these patients. Transvaginal ultrasound recommended every 3–6 months seems currently be the most effective diagnostic tool for the follow-up of young patients treated with cystectomy for benign mucinous cystadenomas.

Laparoscopy in large ovarian tumor is controversial because of concern regarding cyst rupture which might upstage the disease resulting in administration of chemotherapy and also affecting the patients survival if final histopathology shows malignant ovarian tumor. In the case of incidental ovarian cancer, there is always a risk when you are operating for large complex ovarian masses especially in recurrent setting and there are also concerns about trocar insertion site metastasis induced by the chimney effect created by the pneumoperitoneum or direct seeding following contamination of surgical instruments with cancer cells [10–12]. Also, concerns for laparoscopic surgery in patients with large ovarian masses are limited visualization, and technical difficulty of visualizing the ureters and extracting the mass. Ovarian cysts > 10 cm in diameter especially with mainly solid components are often larger than the available laparoscopic endobags [13]. So the practice of laparoscopic surgery in solid ovarian masses more than 10 cm is not recommended.

Conclusions

Mucinous tumors are usually benign and most are multi-locular; management in young patients is challenging, especially in the case of recurrence. Follow-up of these patients is very important, and transvaginal ultrasound seems to be currently the most effective diagnostic tool for the follow-up of young patients treated with cystectomy for borderline mucinous cystadenomas. Total hysterectomy and bilateral salpingo-oophorectomy is recommended after completing her family size or reaching age of 35 for fear of progression and poor compliance in our country.

Compliance with Ethical Standards

Conflict of Interests The author declares that he has no conflicts of interest.

Details of Ethics Approval Approval of the medical ethics committee of the Faculty of Medicine, Alexandria University, was obtained before committing the research and the supporting documents are available on request.

Ethical Approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed Consent Informed consent was obtained from the patient included in the study.

References

- Hart WR (1992) Pathology of malignant and borderline (low malignant potential) epithelial tumors of ovary. In: Coppleson M, Monaghan JM, Morrow CP, Tattersall MHN (eds) *Gynecol Oncol* 2:863–87
- Olesen H, Eismum AK (2001) Recurrent mucinous cystadenoma in an 11-year-old premenarchal girl. *Ugeskr Laeger* 163:6601–6602
- Gotoh T, Hayashi N, Takeda S, Takano M, Kikuchi Y (2004) Synchronous mucinous adenocarcinoma of the endometrium and mucinous cystadenoma of bilateral ovaries presenting during fertility therapy. *Int J Gynecol Cancer* 14(1):169–171. <https://doi.org/10.1111/j.1048-891x.2004.014168.x>
- Baksu B, Akyol A, Davas I, Yazgan A, Ozgul J, Tanik C (2006) Recurrent mucinous cystadenoma in a 20-year-old woman: was hysterectomy inevitable? *J Obstet Gynaecol Res* 32(6):615–618. <https://doi.org/10.1111/j.1447-0756.2006.00453.x>
- Mittal S, Gupta N, Sharma AK, Dadhwal V (2008) Laparoscopic management of a large recurrent benign mucinous cystadenoma of the ovary. *Arch Gynecol Obstet* 277(4):379–380. <https://doi.org/10.1007/s00404-007-0556-5>
- Turkyilmaz E, Korucuoglu U, Kutlusoy F, Efeturk T, Dogan HT, Onan A, Guner H, Taskiran C (2009) Recurrent mucinous cystadenoma: a laparoscopic approach. *Arch Gynecol Obstet* 279(3):387–389. <https://doi.org/10.1007/s00404-008-0718-0>
- Salat-Baroux J, Merviel P, Kuttent F (1996) Management of ovarian cysts. *BMJ* 313:1110–1113
- Taskiran C, Erdem O, Onan A, Bozkurt N, Yaman-Tunc S, Ataoglu O et al (2008) The role of frozen section evaluation in the diagnosis of adnexal mass. *Int J Gynecol Cancer* 18(2):235–240. <https://doi.org/10.1111/j.1525-1438.2007.00995.x>
- Ishioka S, Sagae S, Ito E, Kudo R (2004) Ultrastructural study of benign, low malignant potential, and malignant ovarian tumors. *Med Electron Microsc* 37(1):37–44. <https://doi.org/10.1007/s00795-003-0189-0>
- Ramirez PT, Frumovitz M, Wolf JK, Levenback C (2004) Laparoscopic port-site metastases in patients with gynecological malignancies. *Int J Gynecol Cancer* 14(6):1070–1077. <https://doi.org/10.1111/j.1048-891x.2004.14604.x>
- Huang KG, Wang CJ, Chang TC, Liou JD, Hsueh S, Lai CH, Huang LW (2003) Management of port-site metastasis after laparoscopic surgery for ovarian cancer. *Am J Obstet Gynecol* 189(1):16–21. <https://doi.org/10.1067/mob.2003.330>
- Hopkins MP, von Gruenigen V, Gaich S (2000) Laparoscopic port site implantation with ovarian cancer. *Am J Obstet Gynecol* 182(3):735–736. <https://doi.org/10.1067/mob.2000.103251>
- Goh SM, Yam J, Loh SF, Wong A (2007) Minimal access approach to the management of large ovarian cysts. *Surg Endosc* 21(1):80–83. <https://doi.org/10.1007/s00464-005-0596-5>