



Management of Recurrent Large Bilateral Benign Mucinous Cystadenomas of the Ovary: A Case Report

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Abstract

Background: The recurrence of benign ovarian mucinous cystadenomas is rare, and the presence of these cysts bilaterally is even more uncommon. Overall, 11 cases in our literature review were identified. Of these recurrences, only two cases were bilateral. The management of ovarian cysts in young patients is challenging, especially if they recur. Laparoscopic surgery is the most common intervention for ovarian cysts and a conservative approach using ovarian cystectomy is preferred in women in the reproductive age group.

Case Presentation: A 16-year-old nulliparous female was admitted with persistent lower abdominal pain and bloating. A pelvic magnetic resonance imaging showed large bilateral cystic and multiloculated adnexal masses arising from her ovaries. Her tumor markers were normal. The patient underwent three separate ovarian cystectomies over a seven-year period from the age of sixteen due to recurrent large bilateral benign ovarian mucinous cystadenomas. Repeated histological examinations were the same after each case. Given her age and the history of multiple ovarian surgeries, she was referred to the fertility clinic to explore options for oocyte cryopreservation.

Conclusion: This unusual case of bilateral recurrent benign ovarian mucinous cystadenomas underscores the importance of early laparoscopy and cystectomy as a recommended approach. Postoperative transvaginal ultrasound scans during follow-up may assist in the early detection of recurrence cases. Clinicians should strongly consider referring young patients with a history of repeated adnexal surgeries to infertility treatment centers for fertility preservation.

Keywords: Fertility, Gynaecology, Ovarian mucinous cystadenoma, Reproductive medicine.

To cite this article: Omura M, Sarkar RK, Sankar A. Management of Recurrent Large Bilateral Benign Mucinous Cystadenomas of the Ovary: A Case Report. *J Reprod Infertil.* 2025; 26(1):64-68. <https://doi.org/10.18502/jri.v26i1.18783>.

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Received: 1, Nov. 2024

Accepted: 1, Mar. 2025

Introduction

Mucinous tumors are the second most common type of epithelial ovarian tumors (1). Among them, ovarian mucinous cystadenomas account for 15% of all ovarian tumors (2). They are characteristically unilateral, with only 5–10% of these presenting bilaterally (2). The recurrence of mucinous cystadenomas after complete excision is infrequent, and bilateral recurrence is even less common. Based on our knowledge, there are only two reported cases of bilateral recurrence (1, 3). Ovarian mucinous cystadenomas typically present in women between 30–50 years of age and rarely

occur in children. They can become quite large but 77–87% of all mucinous tumors are benign (4). They typically appear as smooth multilocular cystic structures containing mucoid material on ultrasound (4).

Laparoscopic surgery is now the standard of care in the management of benign ovarian cysts due to lower morbidity and quicker post-operative recovery. This approach is also safe and effective, even with complex or large masses over 10 cm (5). A conservative approach such as an ovarian cystectomy is the preferred method, especially for

young patients who wish to preserve their fertility, since the method minimizes the loss of functional ovarian tissue. However, conserving ovarian tissue can be challenging with large cysts.

This case report describes the management of recurrent large bilateral benign ovarian mucinous cystadenomas in a young patient who underwent three surgeries over a seven year period for bilateral recurrences since the age of sixteen. Furthermore, this case report examines the possible implications for future fertility in a young nulliparous woman who has undergone multiple adnexal surgeries for these tumors. This case report has been written having obtained informed consent from the patient.

Case Presentation

In 2014, a 16-year-old nulliparous women admitted to emergency department (ED) of Barnsley Hospital South Yorkshire United Kingdom (UK) with a one-year history of abdominal pain, bloating, and nausea. Her pregnancy test was negative. She was initially admitted under gastroenterology for further investigations. She had a past medical history of migraines and was on the combined oral contraceptive pill (COCP). She attained menarche at the age of 14 years and had regular periods. Her body mass index was 26 kg/m^2 .

Investigations: A transvaginal ultrasound scan (TVS) revealed a complex cystic structure measuring $8.1 \times 6 \times 4.6 \text{ cm}$, along with a second cyst on the right measuring $12 \times 8.3 \times 5 \text{ cm}$ (Figure 1). Neither ovary was distinguishable from these structures. An urgent gynaecological referral was advised. She was evaluated in the fast track gynaecology clinic and examination revealed a palpable mobile mass in the suprapubic area. A pelvic magnetic resonance imaging demonstrated large bilateral adnexal cysts arising from the ovaries. The cyst relating to the left ovary was multiloculated measuring $7.3 \times 8 \times 8 \text{ cm}$ with thin septations.

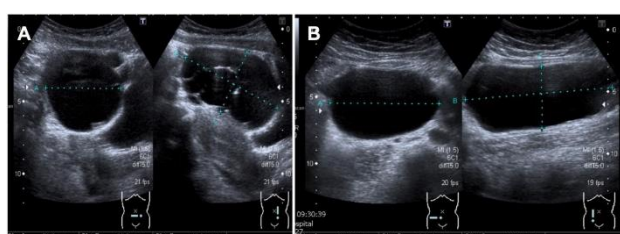


Figure 1. Transvaginal ultrasound images from the first visit demonstrating an $8.1 \times 6 \times 4.6 \text{ cm}$ complex cystic structure in the midline of the pelvis (A) and a second cyst measuring $12 \times 8.3 \times 5 \text{ cm}$ to the right of the midline (B)

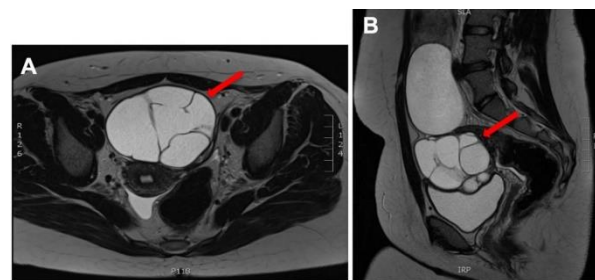


Figure 2. T2-weighted magnetic resonance imaging demonstrating a transverse view (A) and a sagittal view (B) of a multiloculated left ovarian cyst measuring $7.3 \times 8 \times 8 \text{ cm}$ with thin septations distributed throughout

The right ovarian cyst measured $8.6 \times 10 \text{ cm}$ and appeared simple with one septation (Figure 2). There was no evidence of pelvic lymphadenopathy. The tumor markers including CA-125, carcinoembryonic antigen, CA19-9, alpha-fetoprotein, and beta human chorionic gonadotrophin were normal. The case was discussed in the gynaecology multidisciplinary team (MDT) meeting.

Treatment: Following MDT discussion, a plan was made to perform a laparoscopic bilateral ovarian cystectomy with peritoneal washings. Although there were initial attempts to perform the procedure laparoscopically, it was ultimately converted to an open laparotomy. Complete excision of the right ovarian cyst without cyst rupture was performed. This was followed by an ovarian cystectomy on the left side. Histology confirmed bilateral benign mucinous cystadenomas with no evidence of malignancy on cytology from the peritoneal washing. She was discharged back to her primary care provider following a routine follow-up.

During an interim period of five years, the patient was extensively investigated for chronic abdominal pain and was diagnosed with irritable bowel syndrome. During this time period, the patient had four TVS which showed no evidence of recurrence of her bilateral ovarian cysts.

Five years after her initial laparotomy, the patient presented to the ED with right iliac fossa pain and vomiting. A TVS showed a new 5.4 cm right ovarian cyst containing loculations with some solid areas, and a new 5 cm simple left ovarian cyst (Figure 3). Her tumor markers were normal. A laparoscopic right ovarian cystectomy was performed as an emergency procedure (Figure 4). The left ovarian cyst was fixed to the abdominal wall and was not removed due to its position and

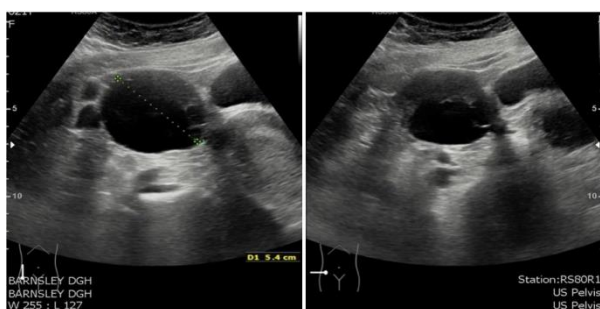


Figure 3. Transvaginal ultrasound images from second visit demonstrating a 5.4 cm right ovarian cyst containing loculations and some solid areas

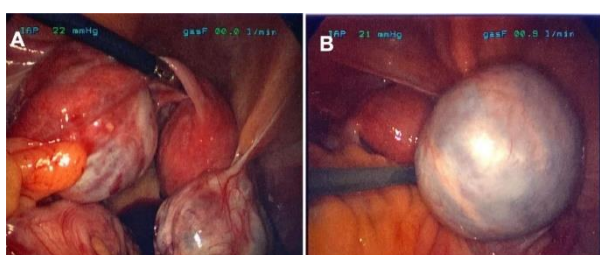


Figure 4. A laparoscopic view of bilateral ovarian cysts at second recurrence (A) with a 6 cm right benign mucinous cystadenoma (B)

smaller size. Histology confirmed the recurrence of a 6 cm right benign mucinous cystadenoma.

Follow-up: Due to the nature of this recurrence, a follow-up TVS was planned. She developed a new 8 cm loculated right ovarian cyst eighteen months later, and the left simple ovarian cyst detected on previous laparoscopy had enlarged to 7.1 cm. Tumor markers remained normal. She subsequently had a laparoscopy with peritoneal washing, adhesiolysis, and bilateral ovarian cystectomy. Both cysts underwent rupture during dissection. The histology confirmed bilateral benign mucinous cystadenomas with no evidence of malignancy on cytology from the peritoneal washing. After discharge, a repeat TVS, conducted twelve months later, did not show any recurrence. Given her age and history of repeated ovarian surgeries, she was referred to a fertility clinic at a tertiary center to discuss oocyte cryopreservation.

Discussion

A literature review indicated that the recurrence of ovarian mucinous cystadenomas is uncommon. Only 11 cases could be identified in the literature available from a first report in 2001 until now (1, 3, 5-11). While recurrence is rare, bilateral recur-

rence in this circumstance is even more unusual. In the 11 cases mentioned above, recurrences were unilateral and located on the same ovary that contained the first excised cyst, and only two cases presented as bilateral recurrences (1,3). The first bilateral recurrent case of ovarian mucinous cystadenoma was published in 2004 which described a 31-year-old lady who had received ovulation induction for primary infertility. She eventually underwent a total abdominal hysterectomy for mucinous adenocarcinoma of the endometrium and bilateral salpingo-oophorectomy after two failed bilateral ovarian cystectomies (3). The second case was published in 2018 and described a 24-year-old lady who underwent open right ovarian cystectomy for a 12 cm ovarian mucinous cystadenoma, followed by laparoscopic bilateral ovarian cystectomy two years later.

A number of factors influence the management of ovarian cysts including patient age, parity, size and nature of the cyst, as well as menopausal status (12). As ovarian mucinous cystadenomas are non-functional, COCP is considered ineffective in preventing cyst formation (8). Laparoscopy is now the standard treatment for ovarian cysts. Laparoscopy offers distinct advantages to open surgery including reduced post-operative analgesia requirements, earlier mobilization, and shorter hospital stays (13). Laparoscopy also has the additional advantage of taking biopsies or washings to rule out malignancy. It allows for the examination of other structures, including the appendix, to assess for differential diagnoses such as pseudomyxoma peritonei, which may also present with multiple mucinous lesions. A concern with the laparoscopic approach is tumor spillage and some controversy exists regarding whether intraoperative cyst rupture is a risk factor for recurrence. Previously, Ben-Ami et al. found ovarian cystectomy and intraoperative cyst rupture to be independent risk factors for recurrence, while Mizrachi et al. suggest that intraoperative cyst rupture does not increase its recurrence rate (5, 11). In our case, there was no spillage during the first operation; therefore, it is probable that intraoperative spillage is unlikely to be the sole cause of cyst recurrence.

Surgical intervention with adequate excision can effectively treat large benign ovarian mucinous cystadenomas; however, there is a possibility of recurrence. The management of ovarian cysts in young patients is challenging, especially with preserving fertility in recurrent cases. A conservative

approach with ovarian cystectomy is preferred in such patients to maximize the preservation of ovarian tissue (9). Laparoscopy should focus on minimizing damage to ovarian reserve by reducing diathermy use and employing hemostatic agents. It is essential to inform patients about the potential effects on fertility and to consider referring them for oocyte cryopreservation. In the UK, there are stringent eligibility criteria for fertility treatments funded by the National Health Service (NHS). UK clinicians should therefore be aware of funding eligibility criteria, as some clinical commissioning groups in certain regions may not cover such cases, necessitating individualized funding. Additionally, the timing of oocyte cryopreservation must be carefully considered. Ovarian stimulation and oocyte collection must be done as an interval procedure when the ovaries are normal to avoid the risk of cystadenoma perforation.

Given the potential for recurrence, follow-up with transvaginal ultrasound surveillance is an effective diagnostic tool and should be considered. Current UK guidelines recommend annual TVS to monitor simple cysts greater than 50 mm in size (14). Therefore, after surgery for recurrent cysts, as illustrated in this case report, a follow-up TVS within 6 to 12 months appears reasonable. This approach facilitates the early detection of recurrences before cysts reach excessive sizes. Clinicians are recommended to take into account features such as size and clinical symptoms to inform their decision regarding frequency and duration of follow-up.

Conclusion

Based on the findings of the paper, recurrent benign mucinous cystadenomas of the ovary are rare, and bilateral recurrences are even less common. This case report will enhance the understanding of the incidence of recurrent large bilateral benign mucinous cystadenomas of the ovary and may inform their management. Early laparoscopy and ovarian cystectomy are recommended, although the techniques can be challenging with larger cysts. TVS may be scheduled for patients following cystectomy if clinically indicated. In young patients, it is essential to take into account their fertility preferences. Moreover, referral to fertility services for the purpose of fertility preservation should be strongly advised.

Conflict of Interest

The authors declare that they have no conflict of interest.

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