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Case Report

Fertility-Sparing Surgery in Infiltrative Mucinous Carcinoma of the Ovary

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Keywords
Fertility · Surgery · Mucinous carcinoma · Infiltrative carcinoma · Expansile tumor

Abstract
Introduction: Mucinous ovarian carcinoma is often diagnosed early and can affect young patients. The preservation of the female reproductive organ is one of the critical issues, especially for nulliparous women. This case report aims to reconsider the safe outcome of fertility-sparing surgery for infiltrative type mucinous ovarian carcinoma. ***Case Presentation:*** A 28-year-old woman with a right mucinous ovarian carcinoma, infiltrative subtype stage IA was treated by right salpingo-oophorectomy, omentectomy, and lymph nodes staging. A 5-year follow-up showed no signs of relapse, and she completed two full-term natural pregnancies. ***Conclusion:*** Conservative surgery is a crucial matter for this patient category. The infiltrative type has a poorer prognosis, but few papers have reported the outcome and the safety of fertility-sparing surgery in this context.

Introduction

Mucinous carcinoma of the ovary (MOC) is a complex epithelial tumor that generally occurs in younger women and is diagnosed early [1]. The latest World Health Organization classification in 2014 to harmonize pathological reporting recommends subdividing mucinous ovarian cancers, either expansile or infiltrative [2].

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MOC is the most frequent subtype for which fertility-sparing surgery (FSS) is considered [3]. Due to the poorer prognosis of the infiltrative subtype, the ovarian cancer clinical practice guidelines recommend removing both ovaries. Therefore, FSS is a big dilemma for young patients in this category [4]. This publication aims to deliver more evidence to the literature regarding the reproductive outcomes and the oncological safety of FSS.

Case Presentation

Method of Research

This case was treated at the Salah Azaiez Institute in 2016. For reference data, we used PubMed using keywords mentioned in page number one. We report a case of a 28-year-old North African woman with no medical or surgical history who consulted first in a general gynecology department for an increase in the size of her abdomen. The general examination found a blood pressure rate of 12/06, with a heartbeat of 69 contractions per minute. The abdominal examination showed an abdominal mass; no inguinal lymph node areas were clinically palpable.

The US revealed the presence of a pelvic tissular mass with cystic areas measuring 12 cm, vascularized by color Doppler. MRI showed a right solid – cystic ovarian mass suspicious of malignancy, measuring 125*80*70 mm. The colonoscopy and esophagus endoscopy were normal. The tumor markers were negative: tumoral antigen (CA 125) and carcinoembryonic antigen.

She underwent surgery, she had a cystectomy, and the frozen section examination confirmed the diagnosis of MOC; the surgery was completed by a right salpingo-oophorectomy, omentectomy, and peritoneal biopsies taken from the diaphragmatic dome, the right and left paracolic gutter, and the pouch of Douglas.

The peritoneal washings returned negative. The final pathology report confirmed the diagnosis of infiltrative mucinous carcinoma, classified IA with CK 20 and 7 positive (Fig 1–3).

Afterward, she was referred to our center. The staff decided to complete the ovarian staging, so she underwent pelvic node sampling on the right pelvic area; the frozen section returned negative, so we completed the left pelvic area without removing the lymph nodes from the periaortic area.

The histopathological study found 11 lymph nodes disease-free (five lymph nodes on the right and six on the left pelvic area). The multidisciplinary meeting decided to monitor the patient; the planned follow-up was based on physical examination and tumor marker every 3 months in the first 3 years and then every 6 months for the next 2 years. The tumor markers were negative during all follow-ups, and there have been no signs of relapse. During the follow-up, two full-term natural pregnancies occurred, respectively, 3 months and 4 years after the end of the treatment (Fig. 4).

Discussion

Through this case report, we observed that, despite the infiltrative subtype, FSS could offer oncologic safety and good reproductive outcomes. Lee and Scully, in 2000, described the expansile and infiltrative types of MOC based on morphological and prognostic differences; based on their growth patterns, the infiltrative type has a poorer prognosis [4].

To the best of our literature review, only two papers were explicitly devoted to the outcomes of FSS in MOC according to tumor histotype specifically. Gouy et al. [5], study in 2018, are the first to examine the outcomes of FSS according to tumor histotype specifically.

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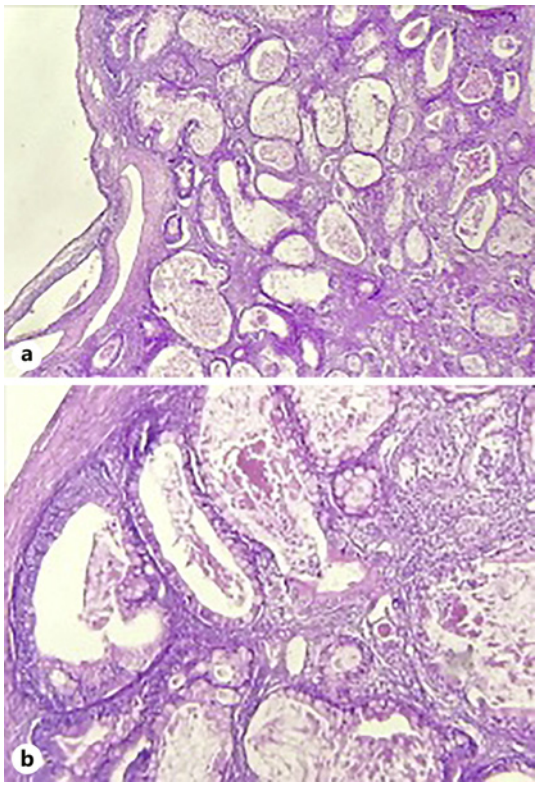

Fig. 1. a, **b** The tumor exhibits infiltrative growth with irregularly shaped glands arranged haphazardly within the altered stroma.

Surprisingly, it showed a low recurrence rate for the infiltrative type in patients who were adequately staged with stage I MOC [6]. As for the fertility results, within 21 patients, there were 6 pregnancies in 4 patients with expansile-type tumors and 2 patients with infiltrative-type tumors. Five pregnancies were spontaneous; 2 patients had pregnancies before the recurrent disease [5].

The second study was conducted in China in 2022 by Wei Lin et al. The study enrolled 159 patients, witch 78 patients had FSS; among them, 12 developed recurrent disease. They reported one death in each group. There was a tendency toward poorer DFS in the infiltrative tumors than the expansile tumors. Twenty-one patients achieved 27 pregnancies, with a live-birth rate of 88.9% [6] (Table 1).

Oncological Safety

Only one paper was explicitly devoted to the outcomes of FSS in MOC, conducted in 2015 by Jung-Lee et al. [7]; the study enrolled 110 patients, 90 patients classified stage I, 35 of whom underwent FSS. The results showed that there was no significant difference in recurrence-free survival or disease-specific survival. Furthermore, the two groups had no difference in recurrence-free or disease-specific survival [6].

The first study mentioned above reported 2 patients recurred out of 21 in each group, 19 and 160 months after the surgery, and they are currently alive [5]. In the second study, the DFS rate was 82.5%, with a 5-year overall survival rate of 98.6% [6].

Lee and Scully reported that 2 patients out of five with infiltrative subtypes who were treated conservatively were disease-free 5 and 12 years later [4]. Rodriguez and Prat described 4 patients out of eleven with infiltrative-type tumors who underwent similar surgeries and were disease-free at a mean follow-up of 5.6 years; the specific outcomes of the 7 other cases were not reported [8].

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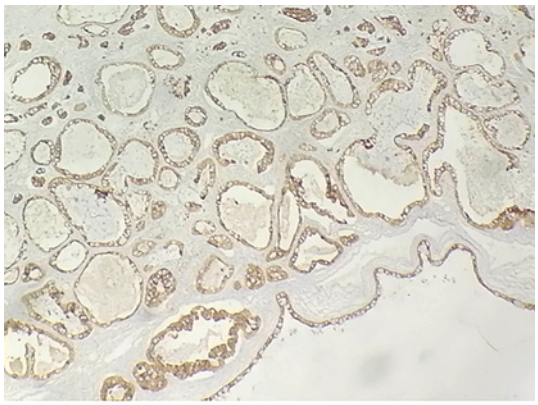

Fig. 2. Tumor cells are diffusely positive for CK7.

Fig. 3. Tumor cells are variably positive for CK20.

Fig. 4. US showing the fetus 9 weeks old after 4 years of the end of treatment.

These different studies highlight the safety of FSS in mucinous epithelial ovarian cancer cases when tumors appear to be grossly confined to the ovaries. However, the criteria for guaranteeing the safety of this procedure remain unidentified and need reflection for selecting certain cases. The CARE Checklist has been completed by the authors for this case report, attached as online supplementary material (for all online suppl. material, see https://doi.org/10.1159/000534737).

Table 1. Summing up the two studies conducted to evaluate the oncological and reproductive outcomes of patients with primary MOC after FSS

Authors	Year of study	FSS	Radical surgery	Recurrent disease	Death	DFS (disease-free survival)	Expansile and infiltrative subtype	conception
Lin et al. [6]	1997–2019	78	81	12 in the group FSS. 6 in the group RS	One death in each group	No significant difference	24: expansile 4: infiltrative tendency of poorer DFS in the infiltrative tumors	21 achieved 27 pregnancies (life birth rate 88.9%)
Gouy et al. [5]	1996–2016	21	---	One in each subtype	0 deaths	160 months	12: expansile 9: infiltrative	6 pregnancies (4: expansile, 2 infiltrative)

Conclusion

In mucinous ovarian cancer, the grading procedure is not like the scale used for serous tumors; the presence of an infiltrative tumor should be considered to discuss FSS. It is a feasible procedure, with possible safe oncological outcomes, but we need more meta-analysis studies with high level evidence to justify using it.

Acknowledgments

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Statement of Ethics

The approval to publish this case report is not required from the Medical and Ethics Committee of the Salah Azaiez Institute, however; this work is done with all due respect to the code of ethics under the supervision of the same committee. This case report was reviewed and the need for approval was waived by the Ethics Committee of Salah Azaiez Institute. Written informed consent was obtained from the patient for publication of the details of their medical case and any accompanying images.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

S.B.L. and M.G.: data collection and drafted the manuscript. Y.Z. and L.H.K.: data collection review of the literature. M.S. and A.C.: a review of the literature and draft of the manuscript. R.C. and T.D.: drafted the manuscript.

Data Availability Statement

The data supporting our findings were taken from the patient's folder; further inquiries can be directed to the corresponding author.

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