

## Conservative Management of Borderline Ovarian Malignancy through Minimal Invasive Surgery: A Case Report and Review of Literature

### ABSTRACT

We present the case of a 27-year-old nulliparous woman with a history of missed abortion and desirous of pregnancy diagnosed with rare borderline mucinous tumor in the left ovary intraoperatively. Considering the borderline tumor status, we made an attempt to preserve the right ovary and thus preserve her fertility using minimal invasive surgical procedure (laparoscopic unilateral salpingo-oophorectomy). The patient subsequently became pregnant spontaneously within 2 months and had a full-term delivery. Therefore, we recommend that carefully and skillfully planned and performed, minimal invasive surgery, if combined with vigilant and regular follow-up, can be an excellent treatment option in the management of patients with borderline ovarian tumors who are desirous of pregnancy and childbearing.

**Key words:** Borderline ovarian malignancy, Laparoscopic oophorectomy, Mucinous tumour

### INTRODUCTION

Ovarian cancer is the leading cause of death from gynecological malignancies. The lifetime risk of ovarian cancer in a woman without any family history of the disease is 1 in 70. Because early ovarian cancer produces few specific symptoms, most women present with advanced stage disease for which the prognosis is poor. The concept of borderline ovarian tumor (BOTs) was first described more than 70 years ago by Taylor,<sup>[1]</sup> but in the past few years, we have actually started understanding the exact biological behavior of these tumors. Earlier due to the clinically intermediate behavior, the nomenclature “semi-malignant” was used. Later, the designation of “BOTs” was adopted by the World Health Organization (WHO) in 1973. BOTs constitute 10–20% of epithelial ovarian malignancies and they pathologically show features associated with malignant tumors but they lack stromal invasion. A lack of stromal invasion leads to a more favorable clinical picture in these patients. In comparison to other ovarian cancers, BOTs tend to be less aggressive and the reason for the same is attributed to its histotypes, low International Federation of Gynecology and Obstetrics (FIGO) stages, and lower frequency of BRCA mutations. Approximately 33–60% serous borderline tumors and 80–90% mucinous borderline tumors are confined to one ovary with only 20–30% extraovarian spread. Therefore, women with Stage 1 borderline tumors have an excellent 5-year survival rate of approximately 95–97%.

BOTs tend to occur in women at approximately 20–40 years of age compared with an average occurrence at 60 years in the case of invasive carcinoma.<sup>[2]</sup>

The risk and protective factors for the occurrence of BOT are similar to those of carcinoma; however, the association with mutations in BCRA genes is exceptional.

Arpita Chakraborty<sup>1</sup>, Nitin Premanand Paidhungat<sup>2</sup>

<sup>1</sup>Department of Obstetrics and Gynecology, Bombay Hospital Institute of Medical Sciences, Mumbai, Maharashtra, India, <sup>2</sup>Department of Obstetrics and Gynecology, Bombay Hospital, Mumbai, Maharashtra, India

#### Corresponding Author:

Dr. Arpita Chakraborty, Department of Obstetrics and Gynecology, Bombay Hospital Institute of Medical Sciences, Mumbai, Maharashtra, India.  
E-mail: arpita.221090@gmail.com

It is found that in 15–30% of cases of BOTs are diagnosed incidentally because they tend to be completely asymptomatic; nevertheless, when there are symptoms, they are often vague and non-specific, such as pelvic pain, abdominal distension, and bloating.<sup>[3,4]</sup>

The classic treatment of choice for patients with BOT seemingly confined to one ovary is conservative in which unilateral salpingo-oophorectomy is performed with surgical staging, which includes exploration of the peritoneal cavity, peritoneal washing cytology, and infracolic omentectomy along with multiple peritoneal biopsies. If bilateral borderline tumors are present, portions of one or both ovaries may be preserved by performing only ovarian cystectomy. It is customary or mandatory to send the dissected specimen for intraoperative frozen section for histological examination.<sup>[5]</sup> Surgical treatment is considered to be non-conservative if bilateral salpingo-oophorectomy is performed. Whatever the surgical approach, reported 5-year survival for Stage I BOTs treated with surgery alone is 95% or even better.

Fertility preservation is always a consideration in the management of patients with BOTs, as the incidence of these

tumors is much higher in the younger age group. The optimal candidate for conservative surgical management is a young patient who has Stage 1A disease and is desirous of future pregnancy. Although BOTs exhibit less aggressive behavior and low malignant potential, fertility-sparing surgery has a recurrence rate of 5–33%.<sup>[6]</sup> The question that remains is deciding about the tumor staging up to which conservative surgery should be considered. Several reports have detailed that conservative surgical approach for future reproductive outcomes can be safely attempted up to FIGO Stage 1C of borderline ovarian cancers.<sup>[7,8]</sup>

Here, we are presenting a rare case of unilateral mucinous BOT which we treated conservatively with minimal access surgery.

## CASE REPORT

A 27-year-old woman presented to us with the complaint of lower abdominal pain for 1 month. Pain was dull aching in nature, mild to moderate in intensity, non-progressive, non-radiating with no aggravating or relieving factors. She was married for 3 years and had a history of missed abortion 2 years back which was medically terminated. Since then, she has been actively trying for pregnancy. Her menstrual cycle has been regular and normal until 2 months back. For 2 months, she has been bleeding every 15 days. She had no history of hirsutism, acne, galactorrhea, discharge per vaginam, dyspareunia, dysmenorrhea, fever, lower urinary tract symptoms, bloating, indigestion, constipation, shortness of breath, and weight loss. Past medical/surgical history was irrelevant to the current problem. There was no history of ovarian, uterine, or breast malignancy in the family. On clinical examination, abdomen was soft. There was no distension nor any guarding, rigidity, or tenderness. There was no palpable lymph node at the inguinal region. Per speculum findings were normal. Per vaginally, there was left forniceal boggy with no cervical motion tenderness or pouch of Douglas nodularity. Her routine blood work was within normal range (hemoglobin = 12 g%, total leukocyte count = 8680/mm<sup>3</sup>, platelets = 169,000/mm<sup>3</sup>, erythrocyte sedimentation rate = 31 min, S. creatinine = 0.43 mg/dl, serum glutamic-oxaloacetic transaminase = 19 mg/dl, serum glutamic pyruvate transaminase = 14 mg/dl, and random blood sugar = 80 mg/dl). Her tumor markers showed marginally elevated carbohydrate antigen (CA)19-9 (CA 19-9 = 36.10, CA 125 = 14.8, alpha fetoprotein = 1.75, lactic dehydrogenase = 154, carcinoembryonic antigen = 1.01, and beta-human chorionic gonadotropin = <12). CT scan showed uterus and right ovary to be normal with left ovary replaced with a cystic SOL measuring 10\*10\*10 cm in the pelvis causing mass effect with displacement of the uterus to the right and adjacent bowel to the periphery.

The patient was posted for operative laparoscopy with frozen section examination. *In situ* findings showed a normal uterus, normal right ovary and tube, left ovary was replaced

with a large cystic mass of approximately 10\*10\*10 cm with the left fallopian tube stretched over the cystic lesion. Rest abdominal cavity had no evidence of ascites, adhesions, omental thickening, or metastatic lesions.

The decision of the left salpingo-oophorectomy was taken and frozen section was sent. Frozen section report showed low malignant potential mucinous cystic tumor. Considering apparent borderline nature of the tumor and young age of the patient and her strong desire of future pregnancy, we decided to go ahead with the fertility sparing surgery. We performed left salpingo-oophorectomy with total omentectomy with pelvic washings. Keeping the fact in mind that the tumor was mucinous in nature, appendicectomy was also performed. The uterus and the unaffected right ovary and fallopian tube were left untouched. The final histopathology confirmed the frozen section report. The patient was discharged with proper counseling and was advised 4 monthly follow-up till 2 years.

Two months post-operative, the patient conceived naturally. The entire antenatal period was uneventful. She delivered a full-term female baby in July 2020. Her post-operative follow-ups have been normal till date with no evidence of mass or cyst in the right ovary.

## DISCUSSION

FIGO classification for BOTs is same as that of other ovarian tumors.<sup>[9]</sup> Almost 80% of BOTs are diagnosed at Stage I, in comparison to 20–30% of invasive ovarian carcinomas.<sup>[10]</sup>

About 50–60% of the BOTs are serous tumors and 30–40% of the total constitutes mucinous type. Grossly, mucinous tumors are larger in size than the serous type. Mucinous tumors are generally multilocular, with fine septa internally, along with intramural nodules.<sup>[10]</sup> Only 10–20% mucinous borderline tumors are complicated with peritoneal implants, and if they are diagnosed, then the presence of pseudomyxoma peritonei must be ruled out. Mucinous BOTs are also divided into intestinal type (80–90%) and endocervical or Müllerian type (10–20%). In the presence of intestinal type, primary intestinal cancer must be ruled out. Endocervical type is often associated with endometriomas or deep pelvic endometriosis.<sup>[3]</sup>

The main obstacle in the management of ovarian tumors in young age group is preservation of fertility. Most women in the younger age group are either nulliparous or have not completed their family at the time of diagnosis. The best part in the management of BOTs is that ultraconservative fertility-sparing method gives high cumulative pregnancy rate. Many studies have shown the calculated pregnancy rate after conservative surgery to vary somewhere between 50% and 90%.<sup>[13–15]</sup>

Our result, in this patient, also supports the feasibility of this type of conservative surgery in the young patient population. In these cases, the decision of unilateral salpingo-oophorectomy, accompanied by the exploration of the peritoneal cavity, omentectomy, peritoneal washing, and multiple peritoneal

biopsies, yields the best treatment results in the form of fertility preservation and future pregnancies. Routine biopsy of the contralateral ovary is not recommended unless gross abnormality is encountered as it increases the chance of future ovarian failure with post-operative adhesions.<sup>[2,10,16]</sup> However, the treatment option of cystectomy, as fertility sparing surgery, is not recommended in case of mucinous BOTs due to the high risk of recurrence in the form of invasive carcinoma.<sup>[17]</sup> It is advised that mucinous BOTs should be carefully examined due to the fact that areas of benign, borderline, and invasive cancers tend to coexist together in these tumors.<sup>[17,18]</sup> For these reasons, mucinous BOTs are associated with a higher mortality rate in comparison to their serous counterpart, and therefore, the treatment of choice should be salpingo-oophorectomy for mucinous BOTs.<sup>[10]</sup>

Another aspect of discussion is the choice between laparotomy and laparoscopic approach for the management of these types of cases. Studies have shown the importance of the type of conservative surgery performed and not the type of approach used (laparoscopy vs. laparotomy).<sup>[19]</sup> Conservative surgery in the form of “only cystectomy” has shown the highest recurrence rate (as high as 35%).<sup>[20]</sup> Even with the laparoscopic approach, a limited surgical staging procedure in the form of exploration of the peritoneal cavity along with washings and targeted biopsies is recommended. Laparoscopic approach has its own advantages as in any other surgery of reduced operative time, trauma, bleeding, post-operative adhesions, and morbidity, along with faster recovery, but the main drawbacks of laparoscopic approach for BOTs include lack of tactile sensation, difficulty in manipulation, and high risk of tumor rupture and intraperitoneal spillage of the cystic content (which can lead to pseudomyxoma peritonei formation in mucinous tumors). Due to the higher risk of intraoperative tumor rupture, tumor size limit of up to 5 cm has been recommended for the laparoscopic approach.<sup>[21]</sup> In our case, though the tumor size was 10 cm × 10 cm × 10 cm, we still went ahead with the laparoscopic approach as fertility preservation was of utmost importance in this case and also because we were confident about our surgical techniques and skills.

There remains a controversy over the treatment protocol of performing resurgery in patients treated with conservative procedure at the time of diagnosis for future pregnancies. Few school of thoughts support the recommendation of directly going for radical surgery as soon as these patients complete their childbearing desires in contrary to others who recommend waiting for recurrence to occur before going directly for radical surgery. It can be debated that radical surgery should be performed sooner not only due to the psychological impact produced by waiting for the relapse to occur but also because of the risk of recurrence in the form of invasive cancer.<sup>[22]</sup>

About 25% of recurrences are diagnosed after 5 years,<sup>[22]</sup> therefore, patients who are treated with conservative surgery should be strictly counseled about the importance of follow-up and close monitoring because of the high rate of relapse. The patients are advised to follow up every 4 months for the first

2 years, then twice a year for at least 5 years and thereafter annually.

Follow-up visits should include clinical examination, transvaginal ultrasound, and CA-125 and CA-19-9 levels, since it appears that some mucinous tumors do not mark CA-125.<sup>[2,3]</sup> CA-125 might not be elevated in early stages of BOTs but its found to be elevated in almost 83% of cases of Stage II to Stage IV disease.<sup>[18]</sup> Transvaginal ultrasound remains the choice of imaging modality for monitoring and may be accompanied by a pelvic MRI.

## CONCLUSIONS

Younger women are more likely to be diagnosed with a BOT than with ovarian carcinoma, therefore, choosing the best treatment for these patients is a real challenge. For women who are yet to complete their family, the treatment modality in the form of conservative surgery through laparoscopic approach, along with very close follow-up, to ensure early diagnosis and treatment of future recurrences, appears to be the best option. On the other hand, women who have already completed their family or those who do not desire to bear children, radical surgery is recommended from the start.

Therefore, in our opinion, we can help the young women diagnosed with BOTs and who are desirous of future pregnancies, through conservative surgery by minimal invasive technique and very close follow-up.

## REFERENCES

1. Park JY, Kim DY, Kim JH, Kim YM, Kim YT, Nam JH. Surgical management of borderline ovarian tumors: The role of fertility-sparing surgery. *Gynecol Oncol* 2009;113:75-82.
2. Tropé CG, Kaern J, Davidson B. Borderline ovarian tumours. *Best Pract Res Clin Obstet Gynaecol* 2012;26:325-36.
3. Lalwani N, Shanbhogue AK, Vikram R, Nagar A, Jagirdar J, Prasad SR. Current update on borderline ovarian neoplasms. *AJR Am J Roentgenol* 2010;194:330-6.
4. Schorge JO. *Cáncer Epitelial de Ovario en Williams Ginecología*. 1<sup>st</sup> ed. México: McGraw-Hill Interamericana; 2009. p. 719-21.
5. Palomba S, Zupi E, Russo T, Falbo A, Del Negro S, Manguso F, *et al*. Comparison of two fertility-sparing approaches for bilateral borderline ovarian tumours: A randomized controlled study. *Hum Reprod* 2007;22:578-85.
6. Palomba S, Falbo A, Del Negro S, Rocca M, Russo T, Cariati F, *et al*. Ultra-conservative fertility-sparing strategy for bilateral borderline ovarian tumours: An 11-year follow-up. *Hum Reprod* 2010;25:1966-72.
7. Battaglia F, Plotti F, Zullo MA, Panici PB, Plotti G. Successful pregnancy after conservative surgery for stage IC ovarian cancer with serous borderline tumor on contralateral ovary: A case report. *Gynecol Oncol* 2006;100:612-4.
8. Swanton A, Bankhead CR, Kehoe S. Pregnancy rates after conservative treatment for borderline ovarian tumours: A systematic review. *Eur J Obstet Gynecol Reprod Biol* 2007;135:3-7.
9. Berek JS, Crumb C, Friedlander M. FIGO cancer report 2012 cancer of the ovary, fallopian tube, and peritoneum. *Suppl Int J*

- Gynecol Obstet 2012;119:S118-29.
10. Fischerova D, Zikan M, Dunder P, Cibula D. Diagnosis, treatment, and follow-up of borderline ovarian tumors. *Oncologist* 2012;17:1515-33.
  11. Trillsch F, Mahner S, Ruetzel J, Harter P, Ewald-Riegler N, Jaenicke F, *et al.* Clinical management of borderline ovarian tumors. *Expert Rev Anticancer Ther* 2010;10:1115-24.
  12. Zapardiel I, Rosenberg P, Peiretti M, Zanagnolo V, Sanguineti F, Aletti G, *et al.* The role of restaging borderline ovarian tumors: Single institution experience and review of the literature. *Gynecol Oncol* 2010;119:274-7.
  13. Lenhard MS, Mitterer S, Kümper C, Stieber P, Mayr D, Ditsch N, *et al.* Long-term follow-up after ovarian borderline tumor: Relapse and survival in a large patient cohort. *Eur J Obstet Gynecol Reprod Biol* 2009;145:189-94.
  14. Yokoyama Y, Moriya T, Takano T, Shoji T, Takahashi O, Nakahara K, *et al.* Clinical outcome and risk factors for recurrence in borderline ovarian tumours. *Br J Cancer* 2006;94:1586-91.
  15. Gotlieb WH, Flikker S, Davidson B, Korach Y, Kopolovic J, Ben-Baruch G. Borderline tumors of the ovary: Fertility treatment, conservative management, and pregnancy outcome. *Cancer* 1998;82:141-6.
  16. Morice P, Uzan C, Gouy S. Cirugía de los tumores epiteliales malignos del ovario. *EMC Ginecol Obstet* 2010;10:1-16.
  17. Koskas M, Uzan C, Gouy S, Pautier P, Lhommé C, Haie-Meder C, *et al.* Fertility determinants after conservative surgery for mucinous borderline tumours of the ovary (excluding peritoneal pseudomyxoma). *Hum Reprod* 2011;26:808-14.
  18. Morice P, Uzan C, Fauvet R, Gouy S, Duvillard P, Darai E. Borderline ovarian tumour: Pathological diagnostic dilemma and risk factors for invasive or lethal recurrence. *Lancet Oncol* 2012;13:e103-15.
  19. Fischerova D, Zikan M, Dunder P, Cibula D. Diagnosis, treatment, and follow-up of borderline ovarian tumors. *Oncologist* 2012;17:1515-33.
  20. Camatte S, Morice P, Atallah D, Thoury A, Pautier P, Lhommé C, *et al.* Castaigne: Clinical outcome after laparoscopic pure management of borderline ovarian tumors: Results of a series of 34 patients. *Ann Oncol* 2004;15:605-9.
  21. Ødegaard E, Staff AC, Langebrenne A, Engh V, Onsrud M. Surgery of borderline tumors of the ovary: Retrospective comparison of short-term outcome after laparoscopy or laparotomy. *Acta Obstet Gynecol Scand* 2007;86:620-6.
  22. Romeo M, Pons F, Barretina P, Radua J. Incomplete staging surgery as a major predictor of relapse of borderline ovarian tumor. *World J Surg Oncol* 2013;11:13.

**How to cite this article:** Chakraborty A, Paidhungat NP. Conservative Management of Borderline Ovarian Malignancy through Minimal Invasive Surgery: A Case Report and Review of Literature. *Bombay Hosp J* 2021;63(2):93-96.

**Source of support:** Nil, **Conflicts of interest:** None

#### Author Queries???

AQ2: Kindly provide running title

AQ6: Please note some reference (11 and 12) are not cited in text part and also duplicate reference (10,19) are found (highlighted).

Please check and cite all references in chronological order

This work is licensed under a Creative Commons Attribution 4.0 International License. The images or other third party material in this article are included in the article's Creative Commons license, unless indicated otherwise in the credit line; if the material is not included under the Creative Commons license, users will need to obtain permission from the license holder to reproduce the material. To view a copy of this license, visit <http://creativecommons.org/licenses/by/4.0/> © Chakraborty A, Paidhungat NP. 2021.