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CASE REPORT

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Fertility-sparing surgery for borderline ovarian Brenner tumor and subsequent childbirth: First case report and a literature review

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Abstract

Borderline ovarian Brenner tumors are rare malignancies mainly diagnosed after menopause. A first pregnancy after fertility-sparing surgery by laparoscopic unilateral salpingo-oophorectomy was reported in a previous article. We now report the first baby birth after unilateral salpingo-oophorectomy for a borderline ovarian Brenner tumor and a review of the literature.

KEYWORDS

Brenner tumor, case report, childbirth, fertility preservation, ovarian neoplasm

1 | INTRODUCTION

Borderline ovarian Brenner tumors (BOBTs) represent <0.01% of all ovarian cancers. Patients with BOBTs are usually asymptomatic, and diagnosis is often accidental. They have an excellent prognosis, and the average age at diagnosis is 50.[1] Few cases are published on fertility-sparing surgery (FSS) on premenopausal women with BOBTs, and to our knowledge, no case of baby birth has been described afterward. We described in a previous article the first case of a spontaneous pregnancy after laparoscopic unilateral salpingo-oophorectomy (USO) for a BOBT, performed in a 40 years old woman, in order to preserve her fertility.[2] We present here the first baby birth after USO for a BOBT and a review on FSS for BOBTs.

2 | CASE REPORT

We reported in 2019 the first and only case of spontaneous pregnancy after a laparoscopic USO for BOBTs (FIGO stage 1A) in a 40 years old woman.[2] The patient got spontaneously pregnant less than 3 months after the surgery, but no information about the follow-up of the pregnancy and its outcome was given on that publication since she was only at the second trimester of pregnancy at that time.

The follow-up of the pregnancy was fine and she gave birth to a baby girl at term by cesarean section, since the patient did not wish a vaginal delivery after a previous cesarean section. The contralateral ovary was normal, and no implant was seen at the examination during the cesarean section (Figure 1). The baby girl was 3020 g at birth and had good Apgar scores (9/10/10).

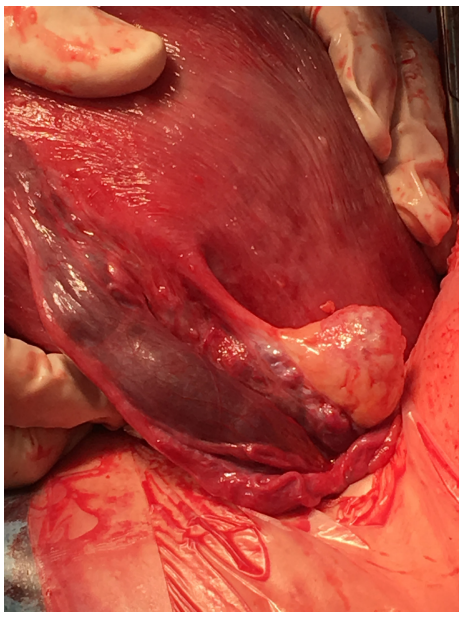

FIGURE 1 Contralateral ovary at C-section

Against what stated in the previous article, neither a hysterectomy nor a controlateral USO were finally performed, according with patient's will. She is using a hormonal intrauterine device as contraception since the delivery. A close clinical and sonographic follow-up every 6–9 months was proposed, even if affected by the pandemic period. The follow-up at 4 years from the first sonographic diagnosis is normal so far.

3 | DISCUSSION

We reviewed three databases (Medline, LILACS, and Central Cochrane Library), and we found seven case series and 15 case reports about BOBTs with a total number of 70 cases.

Table 1 shows the overall BOBTs found in the literature, the cases where the diagnosis was made at or before 40 years old and in these latter women the cases where FSS was performed and a subsequent pregnancy was described. Only two out of the 70 women were treated by USO before or at the age of 40.[2,3]

Cuatrecasas et al.[4] reported a case at the age of 32 where the woman underwent a total abdominal hysterectomy and a bilateral salpingo-oophorectomy (BSO), since the tumor was bilateral and FIGO 2A. Woodruff et al.[5] reported one case out of 10 where the woman had between 31 and 40 years old. However, as the author state, all women in his study underwent a hysterectomy, so no FSS was performed. In Miles et al.[3] series, three premenopausal women were treated by USO, but only the age of one woman is known, being 30. Since in this series of seven women the average age was 50 and the higher age was 64, it is unlikely that the other two premenopausal women were diagnosed before 40 years old. No information about their obstetrical follow-up was given.

Ziadi et al.[6] described the case of a BOBT in a woman of 43 years old which was followed since the age of 30 for a recurrent low-grade papillary urothelial carcinoma. No FSS was performed.

Ricotta et al.[7] recently published 17 cases of BOBTs, with five FSS of which four were by laparoscopy. Interestingly, two women were 81 and one woman was 63 years old. The remaining two women had 43 and 42 years, respectively. No information on follow-up was given for the first woman and the second woman; a BBOT with stromal microinvasion and stage IC1 had a recurrence 3 years later. No information about a possible pregnancy for this latter case was given, even if it seems unlikely.

The good prognosis of BOBTs, as recently shown,[7] supports performing a FSS; however, limitations due to lack of data are evident. In the literature, more studies are available for serous and mucinous borderline tumors (SBOT and MBOT) even if only one randomized control trial exists.[8] Authors suggest that bilateral cystectomy (BC) for bilateral tumors (mainly SBOT) or USO (or even cystectomy) for unilateral tumors can be proposed. Conservative treatment can also be proposed after recurrences according to histology and stage. Recurrence rate is obviously higher in conservative treatment (2.9% for BSO, 12.2% for USO, and 26.7% for cystectomy) but the overall survival does not change.[9,10] Thus, some authors do not endorse radical and/or completion surgery since recurrence rate is 10% after 10 years.[10] Whether FSS could be appropriate also for premenopausal women who had already completed childbearing is also controversial. Commonly follow-up is intensified in the first 2 years, but there is no consensus on how to perform it.[8–10]

We can expect similar results for BOBTs, but data on SBOT and MBOT are probably not sufficient to use the same surgical and clinical strategy. Moreover, the low prevalence of BOBTs and especially of cases with FSS, as shown in Table 1, combined with the lack of strong studies preclude the possibility to make clear and powerful statements about FSS and prognosis for these tumors.[1] Nevertheless, in our case, we opted for a laparoscopic FSS without a completion surgery. The clinical and sonographic follow-up at more than 4 years is normal and has been done every 6–9 months, taking also into account the pregnancy and the pandemic period during the subsequent years. The patient is currently under hormonal intrauterine device. Oral contraception could

TABLE 1 Overall BOBT: cases under 40 years with FSS and subsequent pregnancy

Article	Cases (*n*)	Diagnosis ≤40 years	Fertility-sparing surgery	Pregnancy
Hallgrìmsson et al. (1972)	7	0	—	—
Miles et al. (1972)	7	1[a]	Yes	NK
Woodruff et al. (1981)	10	1	No	—
Svenes et al. (1984)	1	0	—	—
Roth et al. (1985)	6	0	—	—
Hermanns et al. (2000)	1	0	—	—
Cuatrecasas et al. (2009)	7	1	No[b]	—
Uzan et al. (2012)	10	0	—	—
Dierickx et al. (2012)	2	0	—	—
Takahama et al. (2004)	1	0	—	—
Chia et al. (2009)	1	0	—	—
Wang et al. (2010)	1	NK	—	—
Ziadi et al. (2010)	1	0	—	—
Aoun et al. (2014)	1	0	—	—
De Cecio et al. (2014)	1	0	—	—
Klasa et al. (2014)	1	0	—	—
Morales-Palacios et al. (2016)	1	0	—	—
Albu et al. (2016)	1	0	—	—
Garofalo et al. (2019)	1	1	Yes	Yes
Salibay et al. (2021)	1	0	—	—
Ricotta et al. (2021)	17	0	—	—
Yuksel et al. (2022)	1	0	—	—
Total	70	4	2	1

Abbreviation: NK, not known.

[a]Median age 50 (three premenopausal and one at 30 years).

[b]FIGO 2A, bilateral.

probably be a better option; however, no recommendation has been found in the literature on contraception after FSS.

In conclusion, we report the first case of baby birth after a FSS for a BOBTs, without a completion surgery and with a normal follow-up at more than 4 years. The review of the literature found 70 cases of BOBTs with an overall good prognosis, very few cases of FSS and only one case of subsequent spontaneous pregnancy.[2] If the patient wishes so, laparoscopic USO or cystectomy could be proposed, but further studies are needed.

AUTHOR CONTRIBUTIONS

GG involved in study conception, data collection, data analysis, manuscript writing and approval. DB and DT involved in data interpretation, direct patient care, manuscript revision and approval. FB involved in study conception, manuscript revision, supervision and approval.

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CONFLICT OF INTEREST

The authors report no conflict of interest.

DATA AVAILABILITY STATEMENT

Data available on request from the authors.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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