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Case Report

Management of Bilateral Borderline Ovarian Tumor in a 26-year-old Patient in a Resource-Limited Setting

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Abstract

Background: borderline ovarian tumors are rare epithelial tumors, more common before the age of 40, with challenging preoperative diagnosis. Our aim is to describe the management of a case in a resource-limited context in Burkina Faso. *Case report:* a 26-year-old primigravida with a 4-year-old child presented with an abdominal mass evolving over one year in the context of secondary amenorrhea. Clinical examination revealed weight loss, cutaneous-mucosal pallor, increased abdominal volume with tender deep palpation, and an irregular, mildly firm mass around the umbilicus extending to the iliac fossae, with a depressed area along the midline. Speculum examination was normal. On vaginal examination, the cervix was unremarkable, the uterus was of normal size, but the lateral fornices and Douglas pouch were filled. Further investigations revealed microcytic hypochromic anemia and a borderline normal CA 125 level. The rest of the laboratory tests were normal. Abdominal and pelvic ultrasound and CT revealed a strong suspicion of a bilateral ovarian tumour, with no signs of secondary localisation. MRI was not available. The diagnosis of probably malignant bilateral ovarian tumour was retained, and laparotomy was indicated. When the parietal peritoneum was opened, two ovarian tumours with a budding appearance occupied the entire lower half of the abdomen. A part of the omentum was in contact with both masses. Bilateral adnexectomy, total hysterectomy and partial omentectomy were performed in the absence of an extemporaneous histological study. Histological study of the surgical specimens concluded that the tumour was borderline ovarian. Follow-up at 3 years showed no signs of recurrence or tumour extension. *Conclusion*: borderline ovarian tumours are managed surgically, and the choice of conservative or radical surgery is not easy when there is a desire to have children, and when the ovaries are bilaterally involved. Fortunately, the vital prognosis is favourable.

Keywords

Ovary, Borderline Tumour, Abdominal Mass, Amenorrhoea, Burkina Faso

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1. Introduction

Borderline ovarian tumors, are rare epithelial tumors. They represent 10 to 20% of ovarian carcinomas [1]. Their pre-operative diagnosis is difficult. Surgical treatment allows for pathological diagnosis on biopsy specimens or on operative specimens [1-3]. Consequently, the choice between conservative or radical surgery can be challenging for the surgeon, especially in cases where there is a desire for fertility preservation and in the presence of bilateral ovarian involvement [4], particularly without the assistance of extemporaneous histological examination. Our objective is to describe the management of a case of bilateral borderline ovarian tumor in a context of limited resources in northern Burkina Faso.

2. Case Report

A 26-year-old patient was seen in consultation for an abdominal swelling that she had noticed for the past year, in the context of one year of secondary amenorrhea as well. She had a previous pregnancy, childbirth, and was a mother of a 4-year-old child. Her menarche occurred at the age of 13, and her menstrual cycle was regular before the onset of symptoms of the current illness. On clinical examination, the patient presented with weight loss estimated at about 10 kg since the beginning of the symptoms, good hemodynamic status, and cutaneous-mucosal pallor.

The abdomen was enlarged in size, slightly tense. Deep palpation was painful, and revealed an irregular mass around the umbilicus extending to the flanks and left and right iliac fossae. This mass was firm with a depressed area on the midline. Speculum examination revealed a healthy-looking cervix and vaginal walls. There were no bleeding or abnormal discharge observed. On vaginal palpation, the cervix was midline, firm, long, and closed. The uterus was of normal size, but there was slight tenderness upon palpation of the lateral fornices and the Douglas pouch. The rest of the examination was unremarkable.

Blood tests revealed microcytic hypochromic anemia with a hemoglobin level of 7.5 g/dl. The CA 125 level was at the upper limit of normal, at of 35 U/ml. The other blood tests were normal. Abdomino-pelvic ultrasound and computed tomography revealed a strong suspicion of bilateral ovarian tumors, with no signs of secondary localization. Chest X-ray was normal. MRI was not available. The diagnosis of probably malignant bilateral ovarian tumors was made. After transfusion of 2 units of packed red blood cells, pre-anesthetic consultation, a laparotomy via a midline xypho-pubic incision was performed. Upon opening the parietal peritoneum, two budding masses occupying the lower half of the abdomen and corresponding to the two ovaries were visualized. There was no area of normal ovarian parenchyma on these two ovaries. A portion of the greater omentum was in contact with both masses. The uterus, fallopian tubes, and other abdominal viscera appeared normal. There was no intra-abdominal fluid accumulation (Figures 1 and 2).

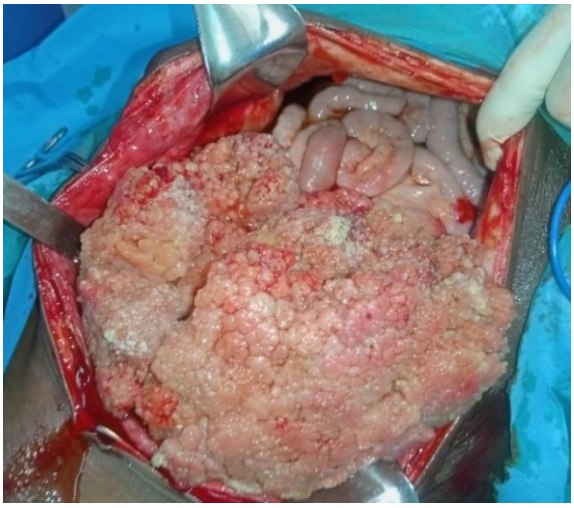

Figure 1. *Appearance of ovarian tumors upon opening of the abdomen.*

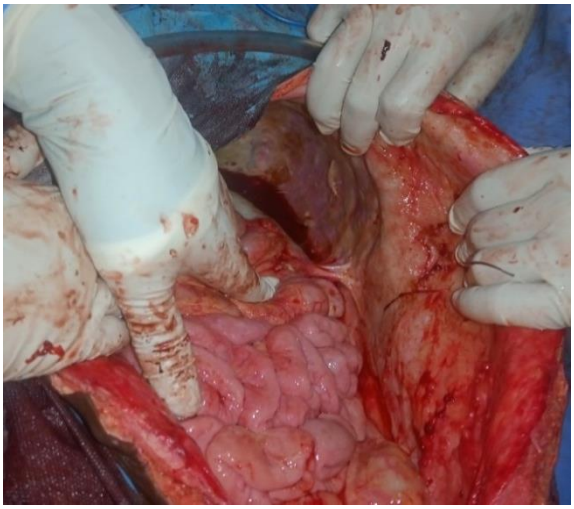

Figure 2. *Normal appearance of the other abdominal viscera.*

After sampling peritoneal fluid, bilateral salpingo-oophorectomy along with total hysterectomy and partial omentectomy were performed. Blood loss was estimated at 100 cc. The abdominal cavity was drained. Postoperative recovery was uneventful. The drain was removed on the second day, and the patient was discharged on the sixth day.

Histological examination revealed, on macroscopic examination, a total hysterectomy specimen of normal size, a partial omentectomy specimen, and the two adnexa, each with a normal tube and abundant budding tissue corresponding to the ovaries. The peritoneal fluid was clear.

Microscopic examination of the two ovarian tumours re-

vealed a proliferation of predominantly micropapillary architecture, consisting of small arborescent papillae lined with moderately atypical multilayered epithelium (figure 3). The other specimens were not invaded by tumour cells. A diagnosis of bilateral ovarian borderline serous tumour was made.

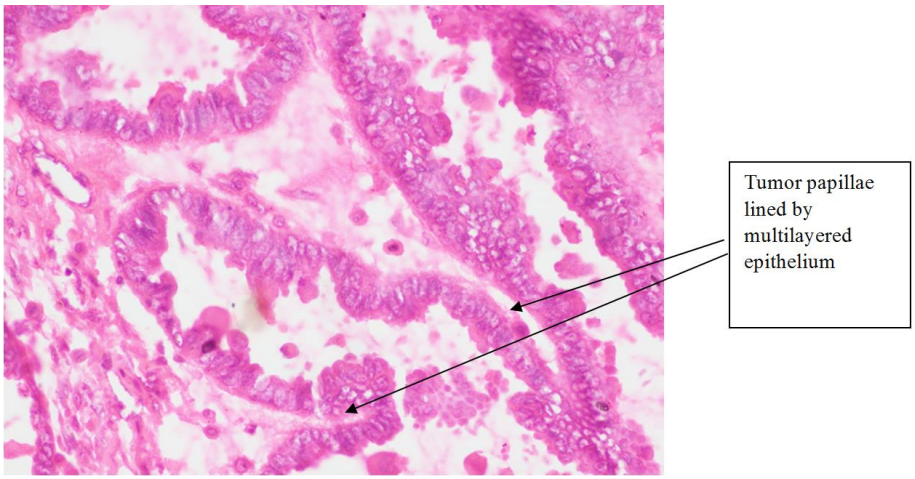

Figure 3. *Serous borderline tumor: papillary proliferation with papillae lined by a multilayered epithelium (Hematoxylin and Eosin stain, 40x magnification).*

The patient was put on an estrogen-progestogen contraceptive pill and was irregularly followed up for 2 years. She did not express any complaints. CA125 levels were not regularly monitored due to lack of resources. CEA and CA 19.9 levels were not available. Ultrasound did not detect any abdominal tumor recurrence during the various follow-ups.

3. Discussion

3.1. Epidemiology

Borderline ovarian tumor is rare with an estimated incidence of 4.8/100,000 per year [3, 5]. We did not find any studies on the subject in Burkina Faso. In the literature, it occurs at a young age, as in the case we present. Indeed, one-third of patients are under 40 years old, and the average age of onset is 10 years younger than that of ovarian carcinomas, whose average age of onset is reported to be 70 years [5, 6].

3.2. Diagnosis

Clinical signs are nonspecific. Those found in our patient were related to a painful abdominopelvic mass frequently described in the literature [2, 6-8]. Secondary amenorrhea helped to guide the diagnosis of the mass towards an ovarian origin, while weight loss raised the hypothesis of a rare malignant ovarian tumor. Indeed, the young age of our patient was not typical of ovarian carcinomas, which typically occur at an average age of 70 years. Malignant ovarian tumors occurring at a young age without signs of virilization are rare [8].

Imaging is also not specific to an ovarian borderline tumor but helps to confirm the ovarian origin of the abdominopelvic tumor. Abdominopelvic ultrasound and computed tomography were important for this purpose in our case. MRI would have allowed for visualization of the macroscopic appearance observed and for staging [5, 6, 9, 10].

The diagnosis of borderline ovarian tumors is based on histological criteria, including four major features: epithelial budding (formation of papillae and vegetations of varying sizes), multilayering of the epithelial lining, increased mitoses, and cytonuclear atypia (moderate). In all cases, there is no invasion of the stroma by tumor cells [8, 10, 11]. Tumor extension into neighboring organs and into the peritoneal fluid helps to characterize the tumor according to the FIGO classification.

These criteria were observed in our patient's samples, and the absence of tumor extension on the omentectomy specimen, on other viscera, and in the peritoneal fluid allowed for classification of the tumor as stage I according to FIGO. Most borderline tumors are discovered at this stage [1, 3, 12]. It was one of the most frequent histological types, serous tumor [6]. This type is bilateral in 25 to 33% of cases, as in the case we describe [1, 6, 8, 12].

3.3. Treatment and Outcome

The treatment of borderline tumors is surgical. The patient in our case was young, a mother of one child, with a desire for future maternity. Conservative treatment would have been the best option. It is acknowledged that conservative treatment is therefore an acceptable option (if not a standard) in a young

patient with stage I borderline ovarian tumor (BOT) compliant with regular follow-up. The only technical limitation to this conservative treatment is the case of stage IB tumor with massive invasion of both ovaries, which does not allow preservation of a portion of macroscopically healthy parenchyma in one of the ovaries. In these cases, bilateral salpingo-oophorectomy should be performed, but with preservation of the uterus and attempt at ovarian cryopreservation [4, 7-9, 13].

The concerning macroscopic appearance of the ovaries in the absence of extemporaneous histological examination in our context led to suspicion of bilateral malignant ovarian tumor in our patient and prompted radical treatment. Elsewhere, under ideal conditions, extemporaneous histological examination is mandatory [5, 8, 9]. It provides reassurance of benign nature and allows for partial ovarian preservation if there is normal ovarian parenchyma and preservation of the uterus in case of desire for maternity [4, 13].

Borderline tumors diagnosed at stage 1 have a good prognosis [8, 9, 14], as evidenced by the case of the patient we present, although biological surveillance means, especially tumor markers, are underutilized in our context. Recurrence seems to be more frequent with ovarian preservation. For stage I, recurrence rates are reported to be 3 to 4% [5, 12]. Malignant transformation would be exceptional, more common in patients over 40 years old [12, 14]. Additionally, due to bilateral ovariectomy, it is important to monitor for signs of early menopause.

4. Conclusion

Ovarian borderline tumors are rare and fortunately diagnosed at an early stage. They occur at a young age, and in our context, they pose challenges in diagnosis and management. The unavailability of extemporaneous histological examination may lead to undue concern about cancer and potentially unnecessary radical surgery, even in cases of future maternity desire. Postoperative care is also marked by insufficient and irregular surveillance, despite the possibility of recurrences and progression to cancer. It is necessary to develop strategies for the management of these rare tumors in our context of resource-limited countries.

Abbreviations

MRI: Magnetic Resonance Imaging
CEA: Carcino-Embryonic Antigen
FIGO: International Federation of Gynecology and Obstetrics

Consent

The patient provided written informed consent for the taking of photographs and their publication in the manuscript.

Authors Contributions

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Ali Ouedraogo: Validation, Writing – review & editing

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Data Availability Statement

All data are already included in this manuscript.

Conflicts Of Interest

The authors declare no conflicts of interest.

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