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Case report

Borderline serous tumor of the ovary discovered during pregnancy: A case report

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ABSTRACT

Introduction: OFTs are tumors with low malignant potential. They represent 10 to 15% of all epithelial tumors of the ovary. Their mean age of occurrence is less than 10 years than that of carcinomas.
Case report: a 29-year-old female patient, second gesture, with no particular pathological history, consulted for pelvic pain occurring during a pregnancy of 8 SA + 5 days. The examination showed an abdominal-pelvic mass lateralized to the left with an enlarged uterus. Abdomino-pelvic ultrasound showed an evolving mono-fetal pregnancy of 10 weeks of amenorrhea, with two right and left latero-cystic solid formations measuring successively 4 × 4.3 cm and 8.99 × 8.25 cm. Pelvic MRI showed a left latero-uterine solid-cystic mass measuring 8.1 × 6.1 × 7 cm. An exploratory laparotomy was performed after the 16th week of amenorrhea revealed a left solid cystic ovarian mass of 10 cm. A left adnexectomy was performed with a right ovarian biopsy, peritoneal biopsy, epiploic biopsy and peritoneal cytology. The pathology report confirmed a borderline serous tumor of the left ovary on the left annexectomy specimen. The right ovary, epiploic and peritoneal biopsy is without tumor proliferation and the peritoneal fluid is acellular.
Discussion: OFT are characterized by their occurrence in women of childbearing age, with an increasingly advanced maternal age for the first pregnancy. Most adnexal masses are diagnosed during the first or second trimester and endovaginal ultrasound in the first trimester is the first-line examination for optimal characterization. Magnetic resonance imaging (MRI) is the recommended second-line examination from 12 weeks of age, in the presence of complex or indeterminate lesions that may be sufficient to distinguish OFMT. Surgical exploration of radiologically highly suspicious adnexal masses during pregnancy is indicated after 15 weeks of amenorrhea, to reduce the risk of miscarriage by alteration of the luteal function of the cyst in the first trimester.
Conclusion: The occurrence of OFT remains rare during pregnancy, which justifies the exploration of any adnexal mass discovered in the peripartum period, for which imaging plays an indispensable role in orienting the diagnosis.

1. Introduction

Frontier tumors of the ovary (OFT) are tumors with low malignant potential. They represent 10–15% of all epithelial tumors of the ovary [1]. The average age of onset is 10 years younger than that of carcinomas. Nearly one third of borderline ovarian tumors occur before the age of 40, in a population wishing to preserve their fertility [2]. The occurrence of OFT during pregnancy is probably related to an advanced maternal age during the first pregnancy, especially in developed countries [3].

The clinical case presented here highlights the interest of radiological, surgical and histological exploration of adnexal masses arising during pregnancy and the route of delivery when OFT is confirmed during pregnancy. This work has been reported with respect to the SCARE 2020 criteria [4].

2. Case report

Patient aged 29 years, 2nd gesture 2nd part, without particular pathological history, she consulted initially for pelvic pain on pregnancy of 8 SA + 5 days, whose examination found an abdomino-pelvic mass lateralized to the left, reaching the flank, poorly limited, slightly

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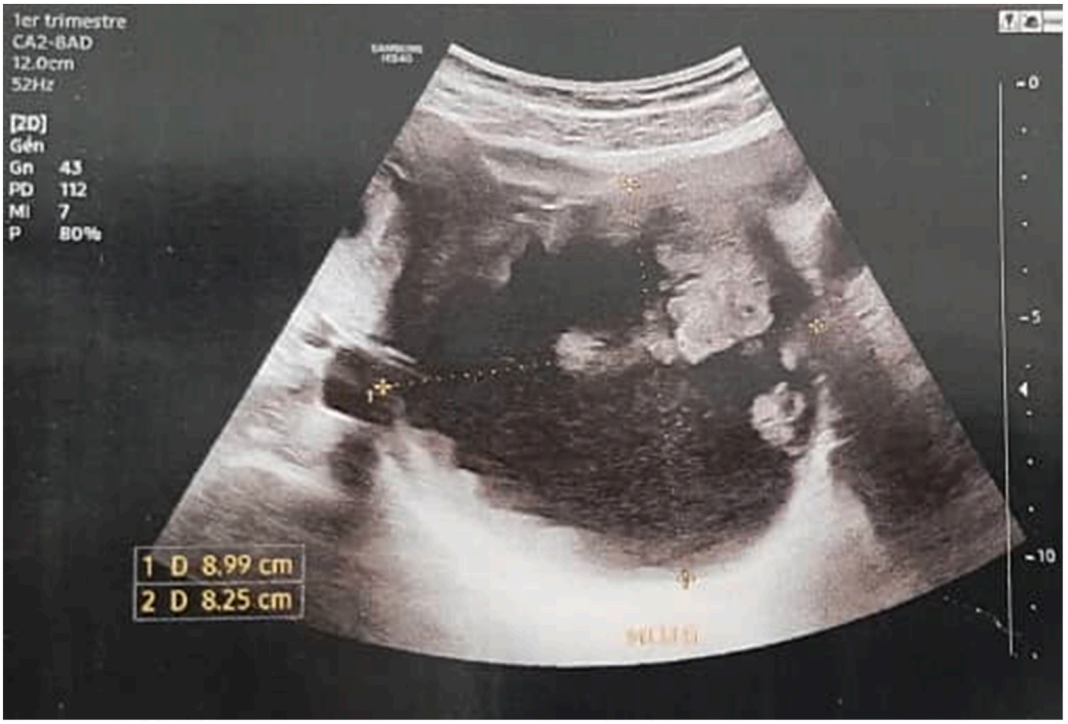

Fig. 1. Left latero-cystic formation with a fleshy part vascularized by color Doppler measuring 8.99 × 8.25 cm in diameter.

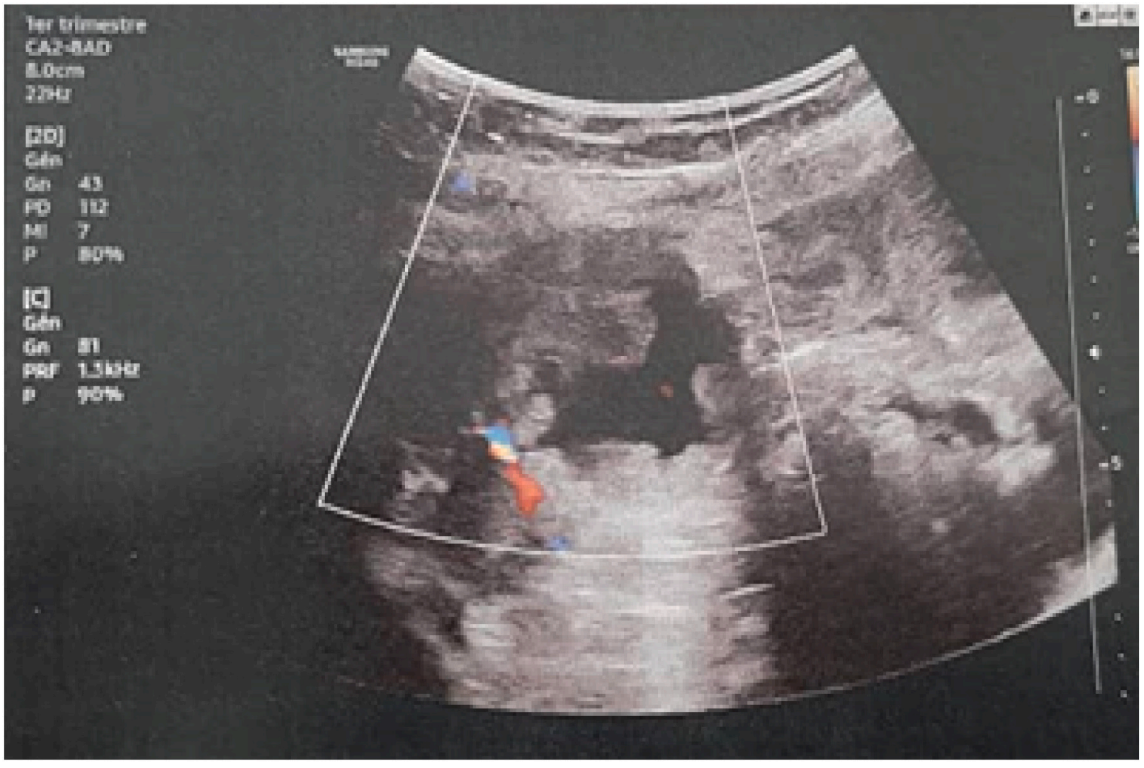

Fig. 2. Right latero-cystic formation measuring 4 × 4.3 cm.

sensitive and a uterus slightly increasing in size.

Abdominal-pelvic ultrasound showed an evolving mono-fetal pregnancy with a cranio-caudal length corresponding to 10 weeks of amenorrhea, with two solid-cystic right and left latero-uterine formations, whose fleshy portion was vascularized by color Doppler, measuring respectively 4 × 4.3 cm on the right (Fig. 1) and 8.99 × 8.25 mm on the left (Fig. 2), with no visible intraperitoneal effusion.

Pelvic MRI performed two weeks after the ultrasound showed a retrouterine mass, lateralized to the left, filling the cul de sac of Douglas, well limited, solid-cystic, continuing with the left ovary, measuring 81 mm × 61 mm extended over 70 mm. The cystic part is of liquid signal, the solid part is of intermediate T1 and T2 signal, with diffusion

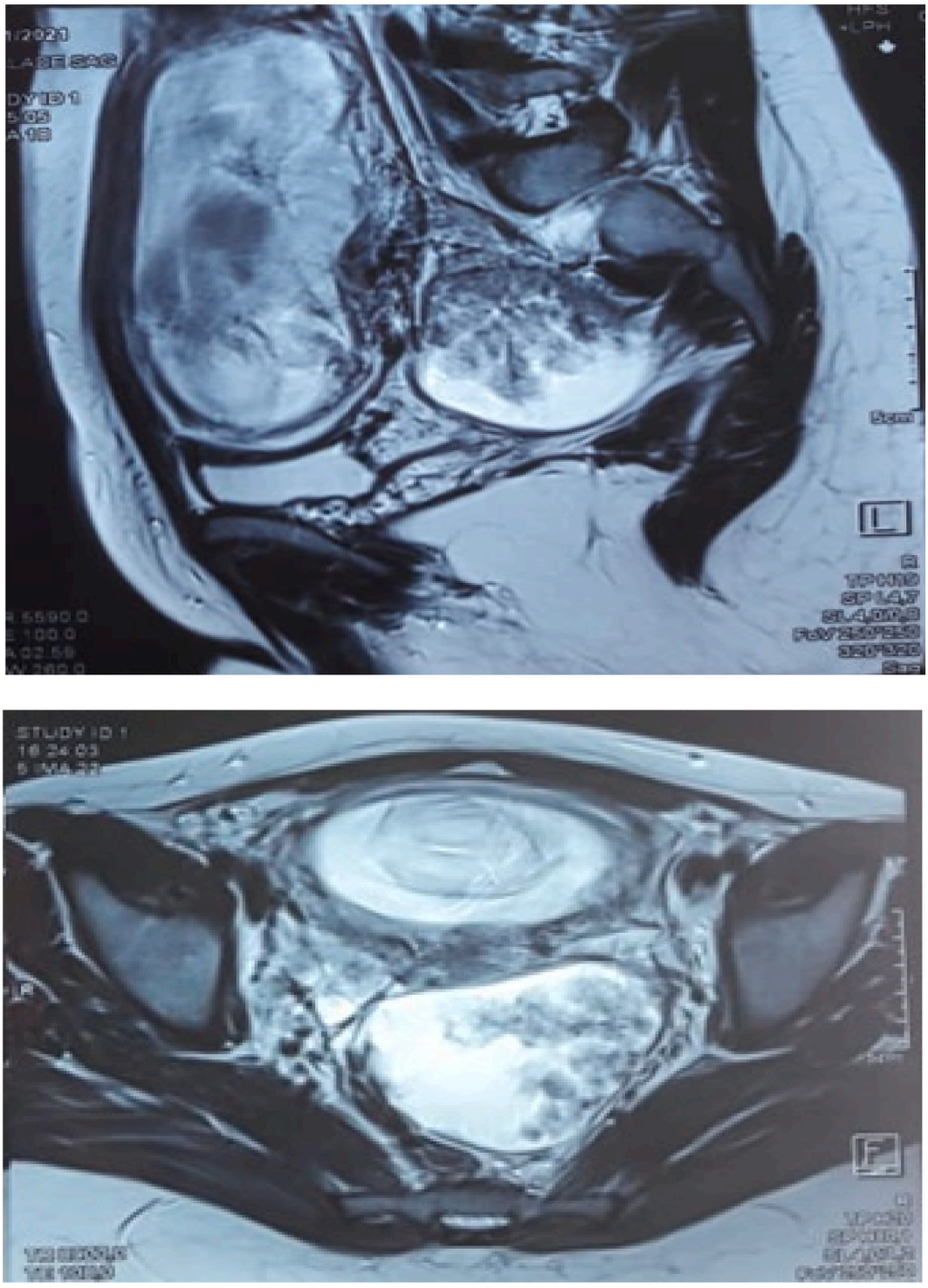

Fig. 3. Retrouterine mass, lateralized to the left, filling the Cul de sac of Douglas, well limited, solid-cystic, continuing with the left ovary, measuring 81 mm × 61 mm extended over 70 mm.

hypersignal (Fig. 3). serum markers, notably CA125: 168 IU/Ml.

The staff decision was exploratory surgery after 16 SA (in order not to alter the functioning of the first trimester luteal cyst and for the placenta to take over hormonally). An exploratory laparotomy was indicated in view of the tumor volume, which revealed a 10 cm long left ovarian mass, depending on the solid-cystic left ovary (Fig. 4) with a slightly enlarged right ovary (Fig. 5). A left adnexectomy was performed with biopsy of the right ovary and staging made of peritoneal biopsy, epiploic biopsy and peritoneal lavage fluid collection. Pathological examination confirmed on the left adnexectomy specimen a borderline serous tumor of the left ovary (Fig. 6) with absence of peritoneal implants. Biopsy of the right ovary, epiploic and peritoneal without tumor proliferation and acellular peritoneal fluid.

The patient was followed after discharge on a regular monthly basis with follow-up ultrasound scans until delivery without recurrence.

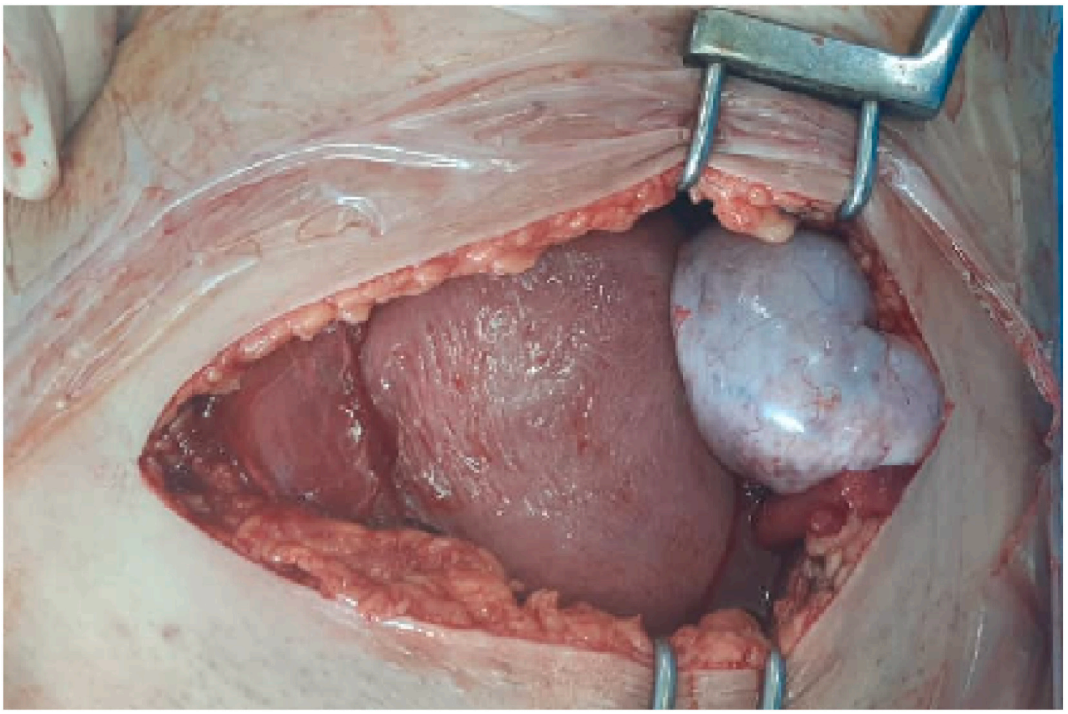

Fig. 4. Left ovarian tumor, 10 cm long axis.

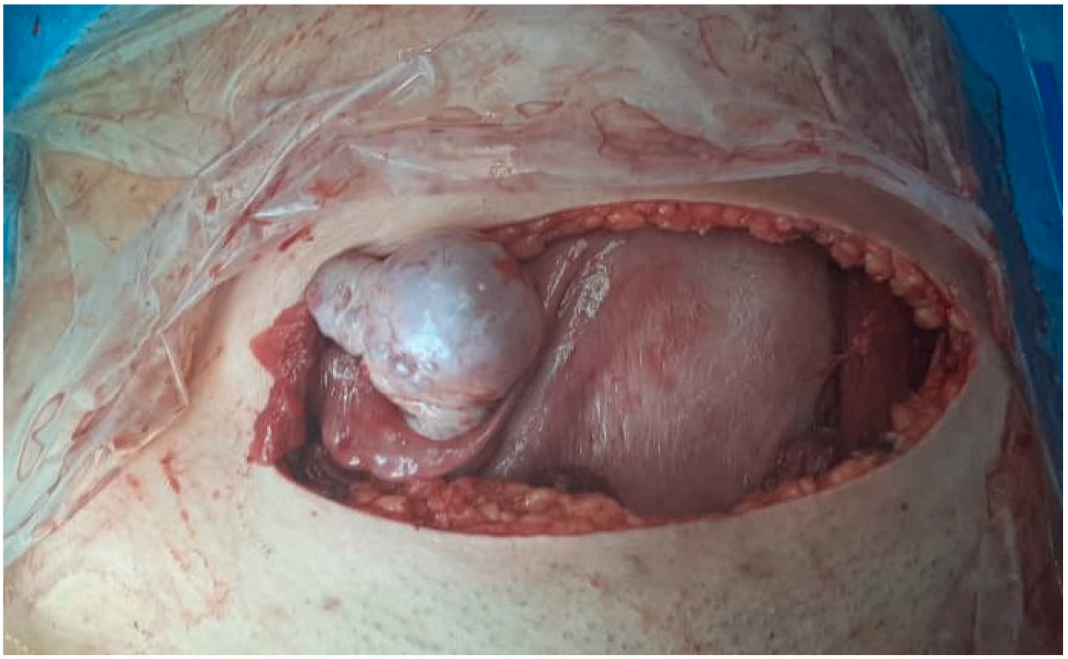

Fig. 5. Right ovary increased in volume.

3. Discussion

Borderline ovarian tumor (BOT) is characterized by its occurrence in women of childbearing age (about 10 years before invasive ovarian tumors) [5], with an increasingly advanced maternal age for the first pregnancy, particularly in developed countries, which justifies systematic screening of any adnexal mass [6]. The prevalence of adnexal masses discovered during pregnancy is estimated to be between 0.2% and 8%, but only 2 to 5% are borderline ovarian tumors [7]. Most adnexal masses are diagnosed in the first or second trimester, so *endo*-vaginal ultrasound in the first trimester is essential for optimal characterization [8]. A study by Timmerman et al. established (simple rules) for endo-vaginal ultrasound in the form of a predictive score (IOTA score) and showed a very high sensitivity and specificity. According to a review of the literature, pelvic ultrasound is the first-line examination for the diagnosis and evaluation of borderline ovarian tumors during pregnancy [9,10], for which the use of color Doppler mode did not find a significant difference [11].

Other complementary examinations will be necessary, in front of complex or undetermined lesions, in order to improve their

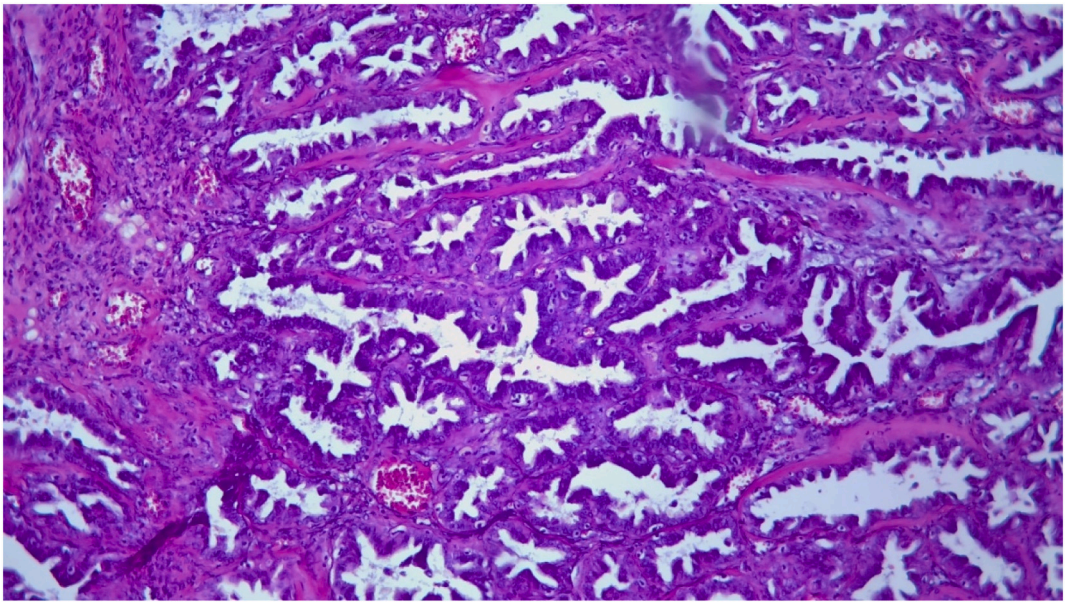

Fig. 6. Large and branched papillae, covered by stratified epithelium of serous type. Absence of stromal invasion (HEx100).

characterization, in particular magnetic resonance imaging (MRI) is the second line examination recommended from 12 SA with T2 and T1 sequences that may be sufficient to distinguish borderline ovarian tumors. Because of the demonstrated fetal risks, the injection of gadolinium should be limited to the maximum. Another study by Thomassin et al. compared the diagnostic performance of pelvic ultrasound with gadolinium-injected MRI using the ADNEX-MR score, which estimates the risk of adnexal lesions with 100% sensitivity [12]. The determination of tumor markers in borderline ovarian tumors suspected during pregnancy has not found its place in a series of studies because of the physiological changes in the maternal body during pregnancy, especially for some onco-fetal antigens (alpha-feto-protein, human chorionic gonadotropin and CA125), involved in feto-placental development. In a series of 40 borderline ovarian tumors occurring during pregnancy, abnormal elevation of CA125 was found, as described in about 40% of OFTs, and normal levels of CA19-9 in all cases [13].

Surgical exploration of radiologically suspicious adnexal masses during pregnancy is indicated after 15 weeks of amenorrhea, to reduce the risk of miscarriage by alteration of the luteal function of the cyst in the first trimester [14], whose approach depends on the volume of the cyst, the risk of intraoperative rupture, and the term of the pregnancy [15]. Minimally invasive surgery by laparoscopy should be preferred because of its safety and feasibility during pregnancy [16]. A preferential use of the laparotomic approach during pregnancy has been described in some studies, especially in case of large tumors, in order to avoid intraoperative tumor rupture. During pregnancy, intraoperative cystic rupture is frequently described in case of laparoscopy (50% of OFT operated) than in case of laparotomy (15 to 20%), so in case of doubt, a laparo-conversion is justified on the feasibility of a conservative procedure without rupturing the mass [17]. The histological nature of OFT is comparable to that found outside pregnancy, with a predominance of serous tumors (55%) as was the case in our observation [18].

Fauvet et al. also studied the histological characteristics of OFT during pregnancy in 40 pregnant patients and described a higher incidence of histological criteria of aggressiveness of OFT when they develop during pregnancy. The micropapillary component is a poor prognostic histologic feature, as it is more often associated with bilateral tumors and the presence of invasive implants. The aggressive character identified during pregnancy, could be explained by the secretion of progesterone and estrogen during pregnancy and the presence of estrogen and progesterone receptors on OFTs. These receptors are most often stimulated during pregnancy [13]. The type of surgical treatment depends on the desire for pregnancy, the bilateral nature of the tumor and the criteria of aggressiveness (peritoneal and extra-peritoneal implants, micro papillary component, invasive) which could explain a unilateral adnexectomy during pregnancy in case of unilateral OFT with staging surgery because several authors mentioned a higher prevalence of OFT diagnosed at an advanced stage in parturients compared to the unaffected population hence the interest of the extemporaneous examination [13].

Surveillance of suspicious adnexal masses of borderline ovarian tumor discovered during pregnancy until delivery is recommended provided that the ovarian lesion has benign features on imaging [15].

Several studies have shown that the route of delivery of patients with a suspected OFT confirmed during pregnancy could be directed by obstetric conditions and not tumor because they have not demonstrated sufficient arguments to know the route of delivery unless the diagnosis of the ovarian tumor was made in the third trimester or a large volume with a risk of rupture in per-partum and without forgetting the tumor location.

4. Conclusion

The occurrence of borderline tumors of the ovary remains rare during pregnancy which justifies the exploration of any adnexal masses discovered in per-partum for which imaging plays an indispensable role to orient the diagnosis. There is not enough data to choose the approach and the type of surgical treatment, which justifies the importance of the multidisciplinary consultation meeting for therapeutic decisions.

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Consent

Written informed consent for publication of their clinical details and/or clinical images was obtained from the patient.

Ethical approval

I declare on my honor that the ethical approval has been exempted

by my establishment.

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CRediT authorship contribution statement

Ely Cheikh TELMOUDI: Corresponding author writing the paper and operating surgeon.
Hamza KIRAM: study concept.
Rajaa ELQASSEH: study concept.
Amine LAMRISSI: correction of the paper and operating surgeon.
Karima FICHTALI: correction of the paper.
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Declaration of competing interest

The authors report no declarations of interest.

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