Case Report

Management Dilemma of an Incidentally Discovered Bilateral Dysgerminoma in a Primigravida During a Caesarean Section: A Case Report

***Adeyemo A,**[1] **Irinyenikan T**[1]

[1] *Department of Obstetrics and Gynaecology, University of Medical Sciences Teaching Hospital, Ondo State, Nigeria.*

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***Correspondence:** Mathew Adeyemo.
Email: matthewadeyemo83@gmail.com

ABSTRACT

Dysgerminoma is a rare malignant ovarian germ cell tumor. It is mostly found among the women of reproductive age but not commonly associated with pregnancy. When it occurs in pregnancy, being a rapidly growing pelvic mass, it can cause fetal malpositioning and presentation, resulting in dysfunctional labour or obstruction. Also, the expected symptoms of dysgerminomas could be unnoticed in pregnancy due to the gravid uterus, only being discovered as an incidental finding causing challenges in its management. It was a case of booked primigravida who had an uneventful antenatal period presented in labour at a gestational age of 39 weeks. The labour was arrested at cervical dilatation of 6 cm with persistent fetal malpositioning. Subsequently, she had a caesarean delivery of a live fetus. Intra-operately, bilateral ovarian dysgerminomas were as found incidentally, however, she desired her future fertility to be preserved. She had unilateral oophorectomy and biopsy taken which was later confirmed to be dysgerminoma on histopathological examination. She was referred to gynaecological oncologist for chemotherapy and follow up. In conclusion, it is possible for dysgerminomas to develop de novo in pregnancy and rapidly grow to form a pelvic mass big enough to cause dysfunctional labour or mechanical obstruction at delivery. Fertility sparing surgery with chemotherapy could be an option in women desirous of future pregnancy if bilateral dysgerminomas are incidentally found at caesarean section.

Keywords: Chemotherapy, Dysgerminoma, Incidental Finding, Fertility Sparing Surgery, Oophorectomy, Primigravida

INTRODUCTION

Ovarian tumors are majorly classified as epithelial, sex cord and germ cell tumors and epithelial cell tumors represents more than 75% of ovarian tumors in general population, however, germ cell tumors are uncommon comprising about 20% of ovarian tumors, both benign and malignant.[1] Meanwhile malignant germ cell tumor of the ovary is rare accounting for less than 5% of ovarian malignancy.[2]

Dysgerminomas are the most common ovarian malignant germ cell tumors and mostly found in women of reproductive age.[3] Its clinical presentations are largely nonspecific, including abdominal pain and swelling. The modality of its management could be either surgical or medical which is fertility sparing.[4] The prognosis is favourable with nearly 100% 5-year survival when detected and treated in early stage of the disease, however, its nonspecific symptoms ofter cause a delay in making diagnosis.[1]

In pregnancy, ovarian malignancy is rare with

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incidence ranging from 1 per 15000 to 1 per 32000 in most reports[2] and dysgerminomas account for approximately 0.2 – 1 per 100,000 pregnancies.[4] Most of the reported cases of dysgerminoma in pregnancy were discovered during the antenatal visit before the onset of labour either as a result of symptomatic manifestations of the tumor or incidental finding from routine antenatal ultrasound scan assessment because its clinical presentations can be masked by the pregnancy symptoms.[3]

In commonly reported cases, incidentally found bilateral ovarian dysgerminoma were managed surgically with bilateral salpingo-ophorectomy.[5,6] This is non-fertility sparing treatment modality which could be considered inappropriate for women who desire future pregnency. Therefore, we present a rare case of bilateral dysgerminoma which complicated the progress of labour and incidentally found at caesarean section in a woman who desired future pregnancy.

CASE PRESENTATION

A 33 year-old booked primigravida who was admitted to the antenatal ward at gestational age of 39 weeks and 4 days on account of intermittent labour pain. She registered for antenatal care at gestational age of 19 weeks and had regular non-adversely eventful antenatal visits. She had three sessions of obstetric ultrasound scan assessment and none was able to diagnose ovarian mass. The palpated fundal height was within the normal limit until the gestational age of 38 weeks when it was noticed to be larger than date and the last transabdominal ultrasound scan was requested to ruled out fetal macrosomia and other abdominopelvic mass. The result came out to be essentially normal findings with estimated fetal weight of 3.3 kg. Patient was rescheduled for further evaluation however, she did not present until when she was in labour.

On admission, patient presented with intermittent labour pain but was yet to rupture the membrane. The fetal heart rate tracing was reactive. Her pack cell volume was 34%. Cervix was 3 cm dilated, soft, anteriorly located. The presenting part was cephalic at station -2. She progressed to active phase of labour after 8 hours and was transferred to labour ward. On getting to the labour ward, she had spontaneous rupture of membrane and was evaluated. The uterine contraction was adequate and fetal heart rate was reactive. The cervical dilatation was 6 cm, fully effaced but station of the presenting part remained – 2. The fetal presentation was assessed to be brow presentation and assessment of cephalopelvic disproportion secondary to brow presentation was made. Therefore, patient was scheduled for emergency caesarean section.

She was delivered a live male neonate by caesarean section, who weighed 3.2kg with good Apgar score. Intraoperatively, bilateral ovarian masses which were large, firm with intact capsule and bosselated surface were incidentally found. The tubes on both sides were stretched over the ovarian masses with no identifiable healthy ovarian tissue. Omentum was essentially normal on systematic inspection and no enlargement of para-aortic or retroperitoneal lymph nodes. There was no suspicious lesion seen on any intra-abdominal organ and no ascitic fluid seen. The patient and her husband were informed but they declared their desire for future fertility. She had left oophorectomy done and biopsy taken. Her intraoperative and postoperative periods were uneventful.

The histology report showed an ovarian mass specimen which weighed 1.0kg and measuring 13 cm in its widest diameter. The cut surface showed reddish solid brown tissue. Microscopically, the cut section showed ovarian tissue with distorted architecture composed of tumour cells which were arranged in diffused sheets, islands and cords separated by scanty fibrous stroma. The tumour cells were uniform in appearance and large with vesicular nuclei and clear cytoplasm. The fibrous stroma generally contained lymphocytic infiltrate and diagnosis of dysgerminoma was made. Post-operatively, the patient was referred to gynaecological oncologist for chemotherapy and follow-up care.

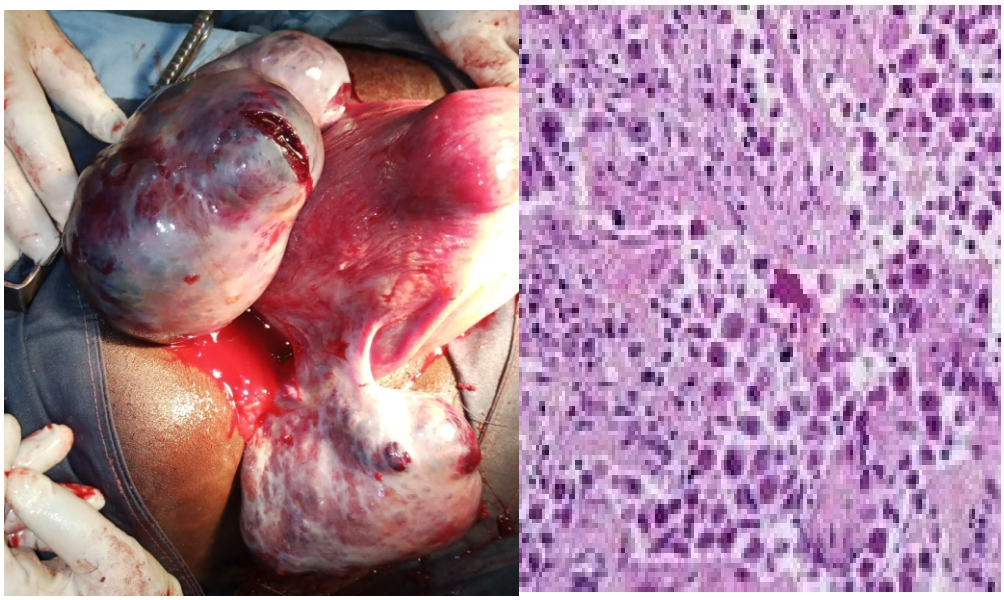

Fig. 1: Bilateral Ovarian Dysgerminomas Intraoperatively and Histology showing sheets of tumor cells separated by fibrous septa, with vesicular nuclei and lymphocytic infiltrate.

DISCUSSION

This is a rare case of incidentally discovered bilateral ovarian dysgerminoma at the time of delivery. She was asymptomatic from the massive ovarian tumor in most part of her antenatal period but during labour she developed poor progress of labour and abnormal presentation most likely due to the effect of the tumor. The cases of dysgerminoma in pregnancy are rarely reported and most of them are unilateral, bilateral dysgerminomas can be found in less than one-quarter of those cases. Germ cell tumor of the ovary is commonly found among women of reproductive age and 3% of which are malignant tumor.[7] Dysgerminomas are a subtype of malignant germ cell tumor and account for about 0.1 to 1 case in 100,000 pregnancies.[8] Therefore, the clinical guidelines regarding the diagnosis and management of this condition in pregnancy are scanty.

In a non-pregnant woman, the symptoms of dysgerminomas could be non-specific and most of them could be observed in women with gravid uterus such as urinary frequency, constipation, pelvic fullness and early satiety.[4] In the presented case, there was no notable symptom she presented with from her bilateral ovarian masses during the course of her antenatal period, probably, they were masked by the normal pregnancy symptoms. The ultrasound examination is crucial in assessing ovarian masses in pregnancy and this is preferably done in the first trimester.[9]

In the case of this patient, the earliest ultrasound scan done was at the early stage of second trimester when she presented to register for antenatal care and there was no definitive impression of ovarian masses. This could be due to the position of the ovaries at the back of the uterus which could easily cause sub-optimal visualization of the ovaries behind the gravid uterus. More so, the uniformly solid texture of the dysgerminoma could make the identification a bit more difficult on sonography compared to a mass with solid-cystic content.[9] There have been documented reports of development of large ovarian germ cell tumors in pregnant women with normal first trimester ultrasound scan.[5,6] Therefore, it is suggested that the dysgerminima in this patient could be hormone sensitive and could have grown exponentially in the late second trimester and the third trimester.

In pregnancy, dysgerminomas like other ovarian masses have increased risk of torsion, rupture and haemorrhage while intrauterine growth restriction has been identified as a common adverse outcome in the fetus.[4] Our case demonstrated none of the above mentioned complications however, there was fetal mal-presentation which resulted into cephalopelvic

disproportion in labour. This could be as a result of dysgerminomas serving as pelvic mass and preventing the fetus from effectively undergoing the mechanism of labour. Therefore this case represented one of the few cases of dysgerminomas reported to be associated with dysfunctional labour.[6]

Generally, suspected ovarian masses in pregnancy are managed by close observation and monitoring with ultrasound and treatment post-delivery if there is no acute complication nor substantial increase in size.[3,10] In a situation where malignant germ cell tumor is diagnosed in pregnancy, conservative surgical treatment should be considered as the initial treatment modality and this has been reported to be preferably done at around 14 – 18 weeks of gestation.[3,4,11] This is to avoid the earlier gestational age which associates with greater risk of spontaneous pregnancy loss if interventional surgery is done at an earlier gestational age with higher risk of adverse feto-maternal outcomes.

In a situation where there is a high index of suspicion for malignancy or huge pelvic mass, vertical midline incision is preferred so as to create enough space for thorough systematic intra-abdominal organs inspection and easy manipulation during surgery.[12] In case of this patient, pfannenstiel skin incision was made because there was no prior diagnosis of a pelvic mass. Unfortunately, this decision limited the complete visualization of the intra-abdominal organs.

In the study done by Zamani et al, on fertility sparing surgery in malignant ovarian germ cell tumor in which they evaluated the reproductive outcome of fertility-sparing surgery and chemotherapy among young women diagnosed of malignant ovarian germ cell tumors of any stage.[13] Out of sample size of 72, 37 of them presented with dysgerminomas and 38 of them were diagnosed at stage III of the disease. Out of this number, 60 patients received adjuvant chemotherapy with bleomycin, etoposide and cisplatin (BEP). The 5 and 10-year disease-free survival rate were 87% and 94.4% respectively. Out of 26 patients who desired future pregnancy, 19 had successful pregnancy that lead to delivery (73%). They concluded that fertility-sparing surgery with unilateral salpingo-oophorectomy could be considered even in patients with advanced stage. In the case presented, the finding was incidental at surgery and the couple were strongly desirous of future fertility, unilateral oophorectomy was therefore done. Postoperative period was uneventful and patient was referred to gynae-oncologist for chemotherapy, close monitoring and follow-up.

It is important to note that dysgerminomas are highly chemosensitive and prognosis is excellent.[12] The patient could be followed-up every 3-4 monthly for 3 years, then 6 monthly for the next 2 years, then yearly up to 12 years.[14] Serum level of Alfa-fetoprotein (AFP) and lactate dehydrogenase (LDH) can be done at follow-up.[11,14] CT scan should be done twice a year particularly if markers are negative at diagnosis. About 97% -100% survival rate has been reported and the fetomaternal outcome of pregnancy with dysgerminomas is good.[13,15]

CONCLUSION

This case showed the possibility of dysgerminomas developing de-novo in pregnancy and growing rapidly to become a large pelvic mass with potential to cause fetal mal-presentation and dysfunctional labour.

Recommendation

If dysgerminomas are incidentally found during caesarean section in a woman desirous of future pregnancy, fertility-sparing surgery can be offered with chemotherapy, and she can be followed up post-operatively with abdomino-pelvic CT scan and tumor markers.

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