Twin Gestation Consisting of Hydatidiform Mole and a Live Fetus: A Case Report

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Abstract

Twin gestation consisting of hydatidiform mole and a live fetus is a rare entity. A 22-year-old gravida 2 para 1+0, 1 alive patient presented with 14 weeks amenorrhoea and vaginal bleeding. Ultrasonography revealed hydatidiform mole and a viable fetus. She had suction curettage at 16weeks gestational age because of heavy vaginal bleeding.

Histology confirmed benign hydatidiform mole. Urinary HCG level declined progressively and was negative by 4 weeks post-evacuation. Eight months after, she was 20 weeks gravid. She had uneventful pregnancy, labour and delivery of a baby boy. The baby (now a year old) and the mother remain well.

Keywords: Hydatidiform Mole, Single Fetus, Twin Gestation, Ultrasonography

Introduction

Hydatidiform mole (HM) with a coexistent of live fetus is a rare occurrence. The incidence is about 1 in 10 000 to 1 in 100 000 pregnancies[1,2]. Molar pregnancies result from the abnormal contribution of genetic material from the egg and sperm[1-5]. Traditionally, there are two possible conditions: a partial mole with an abnormal triploid fetus and a complete mole combined with a normal fetus and placenta[3]. Presentations commonly include first or second trimester vaginal bleeding, rapid uterine enlargement, excessive uterine size for dates, hyperemesis gravidarum or preclampsia before 24 weeks[4].

Ultrasonography is a vital diagnostic tool in this condition and has a role to guide its evacuation[4-7]. Recently, MRI has been used for prenatal diagnosis of molar pregnancy [8]. Other important diagnostic tools include urinary and serum human chorionic gonadotrophin (hCG) titre, DNA typing and histopathology[3].

Nigeria, with approximately 1 in every 22-35 deliveries as twins, has one of the highest twinning rate in the world [9]. However, as far as we are aware only two cases of molar gestation coexisting with a live foetus have been reported in Nigeria [10, 11], with both from the south eastern zone, implying also the rarity of this condition in Nigeria.

We present a case of twin gestation consisting of an HM and a live fetus in a Nigerian woman diagnosed at 12weeks gestation during routine ultrasonography at the fetal monitoring unit of University of Ilorin Teaching Hospital, Nigeria. Pregnancy had to be terminated abruptly at 16weeks gestation because of uncontrollable antepartum haemorrhage.

Case Report

A 22 yr old, Gravida 2 para 1 + ^{O} presented

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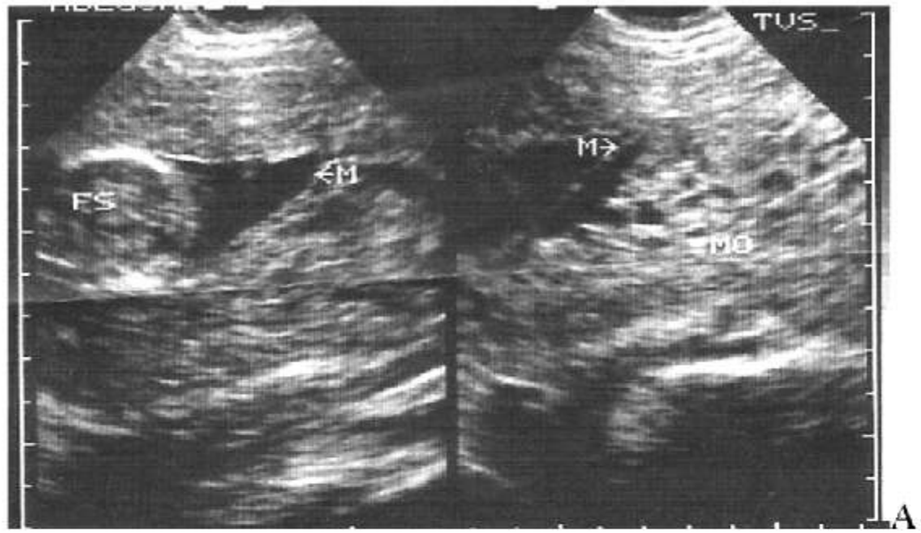
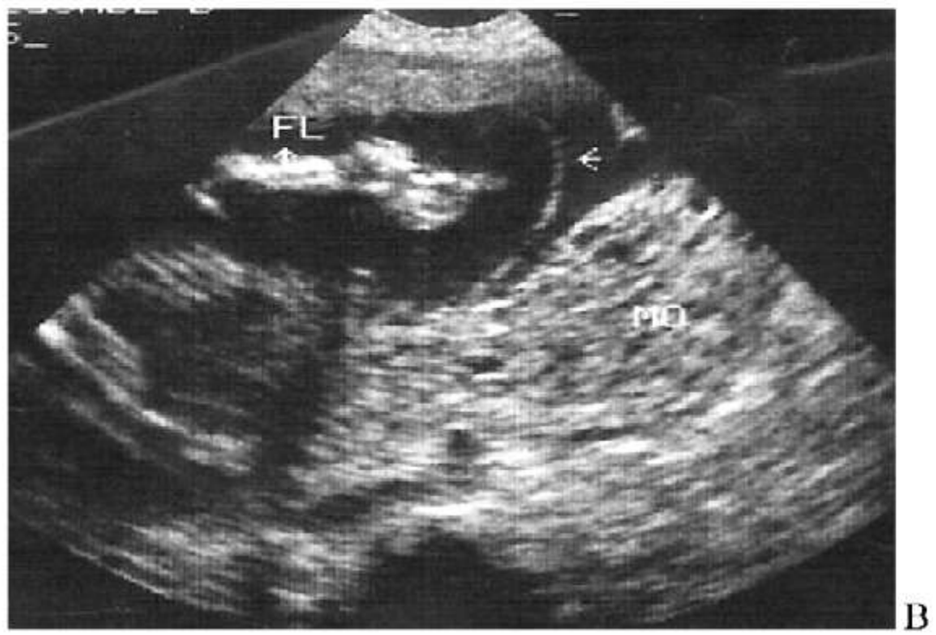
Fig 1a & b; showing twin gestation consisting of a live fetus at 14weeks gestation and a complete mole. Note the fetal skull (FS), femur length (FL), molar tissue (MO) and amniotic membrane (M).





with intermittent spotting of blood of one-week duration. She has been amenorrhoiec for 14 weeks prior to presentation. Physical examination revealed fundal height of 18 weeks duration in a normotensive woman. Ultrasound examination revealed two sacs (fig.1). The leading sac in the lower uterine segment showed mixed echogenic area with intervening cysts giving a 'snow storm' appearance which is characteristic of molar pregnancy. The second sac contained a live fetus of 14weeks gestational age with

regular cardiac activity, adequate amniotic fluid volume and anteriorly situated placenta in the body of the uterus. A thin membrane separated these sacs suggesting diamniotic twins (Figures 1a and 1b). Laboratory work up shows raised serial urinary human chorionic gonadotrpin (hCG) titre of about 2, 000, 000 IU/L. full blood count and LFT were normal. Pack cell volume (PCV) was 25%.

A week later, patient was admitted due to worsened intermittent vaginal bleeding. PCV had fallen to 21%. Repeated US still confirmed a live fetus. Pregnancy was therapeutically terminated at 16weeks of gestation via suction evacuation because of severe anaemia from uncontrollable ante partum haemorrhage. Histological examination showed benign hydatidiform mole. Patient remained stable and serial urine hCG level progressively diminished overtime and became negative from 4 weeks post-evacuation. Chest radiograph post-up and abdominopelvic ultrasound were normal. Eight months after evacuation, she was confirmed to be 20weeks gravid by ultrasound. She had an uneventful antenatal period, labour and delivery. Presently the patient is stable, and both mother and baby are well one year after delivery.

Discussion

Gestational trophoblastic disease of pregnancy otherwise known as HM can be separated into two entities with respect to cytogenetics, histopathology and morphology[3, 4, 12]. Firstly complete HM, which consists entirely of malformed placental tissues forming grape-like, clusters. The genetic component in a complete mole is diploid and is entirely of paternally inherited chromosome. About 90% are 46XX, theorized to originate from a single sperm fertilizing a nullisomic egg followed by duplication of all chromosomes. However, a small percentage of heterozygous 46XX or XY karyotype believed to arise when 2 sperms fertilize a nullisomic egg can also occur[3, 4, 12].

Secondly, partial mole with ascertainable embryo, umbilical cord or an amniotic membrane and consists only focal changes of placenta villi and trophoblast. It has a triploid karoytype, usually from dispermy leading to 23 maternal and 46 heterozygous paternal chromosomes[3, 4, 12].
Therefore, HM with a coexisting fetus can be established by the partial mole syndrome or by a twin pregnancy where the other conceptus has degenerated into a mole[3].

HM is generally a disease of a fertile woman of reproductive age group as in this case presentation. However, cases of molar pregnancy occurring in postmenopausal woman have been documented. Ozumba et al [10] reported a case of HM co-existing with normal pregnancy in an apparently menopausal 56-year-old woman in Southeastern Nigeria. Garcia et al[13] also reported molar gestation in a 61-year-ld post-menopausal woman in the United States of America.

Prenatal diagnosis of coexisting mole and fetus can depend upon the clinical symptoms, signs, physical examination sonographic findings and abnormal biochemical data and cytogenetic analysis. Patients with molar pregnancy, most commonly present in the first or second trimester with vaginal bleeding, rapid uterine enlargement, and excessive uterine size for dates, hyperemesis gravidarum or preclampsia before 24 weeks [4]. Hyperthyroidism and Pregnancy Induced hypertension have been observed in some cases of molar gestation[5, 14]. However, these conditions may not always be present, our patient was normotensive and had no hyperemesis but presented with vaginal spotting and big for date uterus.

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An early and correct diagnosis is imperative to plan subsequent management of such patients [5-7]. Ultrasound plays the most important role in the diagnosis of this condition [4-7]. The typical ultrasonographic findings of a complete molar pregnancy consist of an enlarged uterus,echogenic regions representing molar tissue, and uniformly distributed cystic spaces ranging from few mm to 30mm which are due to the hydrophilic villi seen within the molar tissue given a snowstorm appearance [4]. This should be differentiated from a missed abortion with its degenerated gestational sac, especially during the early pregnancy, or a partially necrotic leiomyoma which can produce a similar appearance[4, 5, 15].

Ultrasound also allows the number of fetuses, placentae and site of placental implantations to be determined. Most importantly it helps to determine presence of foetal anomalies and viability. In addition to being the best method for the diagnosis of HM, ultrasound is also used in the surgical treatment of the mole by suction evacuation under ultrasonic guidance.[6] Recently MRI is being used in some centers [8]. However, its high cost and relative unavailability may limit it use in the developing world.

In this patient, ultrasound revealed a normally appearing foetus with good cardiac activity and a normal anteriorly located placental. The second sac devoid of a fetus, consists of placenta tissues composed of complex cysts with intervening echogenic septa suggestive of HM. HM co-existing with live fetus or fetuses generally presents a management dilemma between clinicians and parent on whether to continue or terminate pregnancy immediately. At present there are limited data to guide the antenatal management of twin pregnancy consisting of HM and a coexisting foetus [4]. However, many clinicians have advocated that Patients who desire to continue pregnancy after such a diagnosis must be cautioned about the potential for severe medical complication like heavy vaginal bleeding and pre-eclampsia which usually warrants termination or pregnancy[4]. In this case presentation pregnancy was terminated on account of uncontrollable heavy vaginal bleeding and moderate anemia. More-so patients should be advised of the high risk for developing gestational trophoblastic neoplasia . A one in four chance of live birth, about 35% risk to develop persistent trophoblastic disease after delivery and at least 20% risk of an early onset of pre-eclampsia have been reported in association with molar pregnancy[15]. In addition such women have a 29% risk of fetal loss due to late miscarriage, intrauterine death and neonatal death[15].

The treatment of this condition is evacuation of the uterus, and vacuum aspiration is considered the method of choice, as opposed to medical induction using oxytoxic drug, which is thought to carry an increased risk of persistent trophoblastic disease [7, 11]. A procedure of the evacuation of complete HM by vacuum suction and continuous ultrasonic monitoring has also been advocated [6]. Because of the possibility of progression to malgnant trophoblastic disease, careful and prolonged follow-up of such women is required. In this case presentation, Urinary HCG level was undetectable 4 weeks after evacuation. Eight months after evacuation, she was 20 weeks gravid. She had uneventful pregnancy, labour and delivery of a baby boy. The baby (now a year old) and the mother remain well.

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