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Case Report

Maternal Klippel - Trenaunay syndrome with complete hydatidiform mole and coexistent fetus: A case managed by surgical evacuation with review of literature

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Abstract

Introduction: Klippel-Trenaunay Syndrome (KTS) is a rare triad of congenital vascular malformations involving extensive Port wine stains, soft tissue or bone hypertrophy and underlying venous and/or lymphatic malformation affecting limb, pelvic or abdominal organs. Pregnancies with a hydatidiform mole and a live fetus are extremely rare, arising in about 1 in 20,000 to 100,000 pregnancies.

Aim: We report a case of a pregnant woman with KTS with twin pregnancy (one fetus and a coexistent mole) with its management dilemma.

Patient: A 27 years old patient, G2P1, NVD, was admitted in our hospital with bleeding per vaginum. Uterus was 24 weeks. USG showed single live intrauterine fetus with 14 weeks gestational age with normal placenta with another placenta with molar changes with adnexa free. β HCG level on admission was 220,000 mlu/ml.

Results: After thorough counseling, the pregnancy was terminated by patient's desire. Induction of abortion with misoprostol was done but severe bleeding occurred mandated surgical evacuation with vaccum aspiration. Histopathological examination confirmed vesicular mole with normal fetus. Her β hCG reached normal levels at the end of five weeks, and she is now on post-molar surveillance for one year.

Conclusion: Termination of pregnancy in twins with coexistent mole is recommended especially far from age of viability. KTS and molar pregnancy may be associated as both are vascular abnormalities that need further evaluation.

Key words: Klippel-Trenaunay Syndrome; pregnancy, twin; ultrasound; complete mole; termination; surgical evacuation

Introduction

Klippel-Trenaunay syndrome (KTS) is characterized by capillary malformations, cutaneous vascular nevi, bony or soft tissue hypertrophy, and abnormal deep or superficial veins. Varicose veins usually affect limbs but canalso be present in abdominal or in-

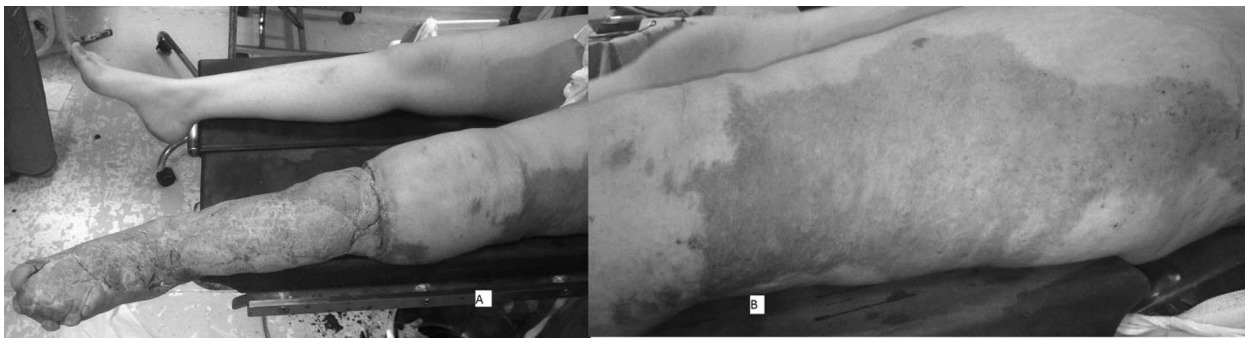

Figure 1: Klippel-Trenaunay syndrome with venous malformations,and hypertrophy ofinvolved left lower limb, lymphedema was present that was managed by debulking surgery with hyperkeratosis in the foot and leg with vascular malformation in the thigh

trapelvic organs. The morbidity of this disease is related to vascular anomalies, which can end in venous insufficiency, thrombophlebitis, limb disparity and thromboembolic disease. The KTS occurs sporadically although it has sometimes appeared in more than one member of the same family. KTS incidence during pregnancy is unknown but it is extremely rare. Pregnancy can increase complications, mainly thromboembolic and haemorrhagic events[1-4].

A multiple pregnancy with one fetus and a coexisting hydatidiform mole has an incidence of 1 in 22,000 to 100,000 pregnancies[5], with most being complete hydatidiform moles (CHM) with a fetus; however, the reported prevalence for a partial mole with a coexisting fetus is 0.005.0.01% of pregnancies[6]. Very few twin pregnancies with a hydatidiform mole and a fetus continue to term as they often have spontaneous or induced terminations for maternal complications.

We present a case of a pregnant woman with KTS with complete hydatidiform mole and coexistent fetus that was detected early in pregnancy. This is the first case to report this association. This case deserves reporting because of rare asscoaition. Also, we discuss the management dilemma with review of literature .

Case Report

We reported a case of 27-year-old woman, G2P1, NVD, with a female living child about 5 years old. The patient suffered KTS since her childhood with left leg affected. The patient had debulking surgery at 6 years old wearing permanent elastic compression since then. There was no family history of this disease in family. Menarche occurred at 12 years with regular menstrual cycles since then. No hypotension or loss of consciousness happened when changing the elastic compressionor while standing up.

She was referred to our hospital at 14 weeks of gestation with the report of twin gestational sacs: One with a live fetus and the other was a complete hydatidiform mole. The patient did not receive any ovulation induction for this pregnancy. Her general and systemic examinations were unremarkable except for the KTS affecting left lower limb with disparity between limbs regarding size, hyperkeratosis from previous surgery from the leg down and vascular malformation over the thigh (Figure 1). The uterus was found to be at approximately 24 weeks of gestation. A transabdominal scan (TAS) at our hospital confirmed the findings of a live fetus with a normally appearing placenta in one sac and an adjacent mixed echoic mass with a honeycomb-like pattern, suggestive of a hydatidiform mole (Figure 2). The fetus was situated over the cervical os, while the molar tissue was situated near the fundus. No bilateral theca lutein cysts were found. The beta human chorionic gonadotropin was very high, at 220,000 mIU/mL; however, her thyroid function tests were normal. Hemoglobin level was 12 gm%. She was counselled regarding the risks involved in continuing the pregnancy, and as per her prefer-

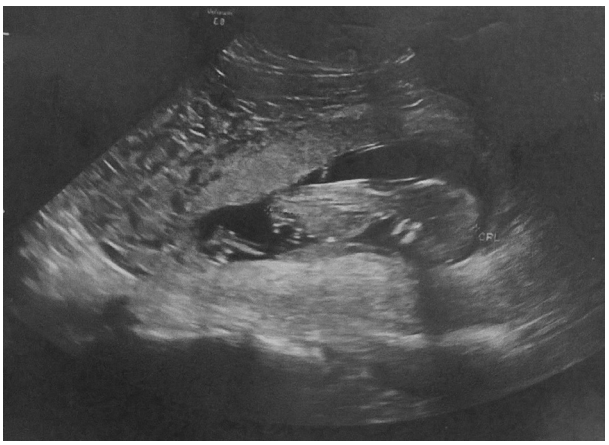

Figure 2: Trans abdominal ultrasound showing upper molar mass near the fundus beside a normal placenta and lower fetus

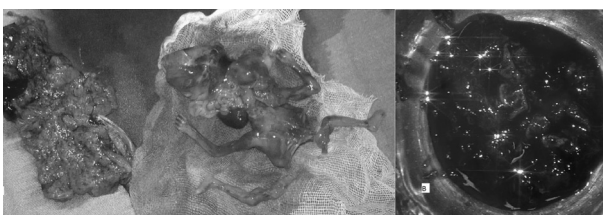

Figure 3: Normal fetus and placenta evacuated by extraction (a) and vacuum aspiration of the molar changes (b)

ence, the pregnancy was terminated. The patient was given oral misoprostol every 4 hours for expulsion of the fetus as it was the lower sac and 10 cm fetal size avoiding manipulation of the fetus with evacuation. Trophoblast embolization is a theortical risk and its risk in this case is lower regarding the risk of fetal extraction. Heavy vaginal bleeding with ballooned cervix and severe pain occurred, so she was transferred to OR . Surgical evacuation under general anaesthtesia and endotracheal intubation where the cervix was dilated by Hegar`s dialtor, evacuation by ovum forceps for the fetus then vaccum aspiration for the complete mole while oxytocin infusion was given (Figure 3). Nothing was abnormal on ultrasound regarding Doppler study and bleeding was not excessive, so evacuation was done under ultrasound guidance to avoid fundal perforation especailly on the thinned right side of the uterus (molar tissue site). During surgery, there was no complications and no problems in the postabortive period regarding abnormal bleeding. Histopathological examination confirmed vesicular mole with a normal fetus. Serial beta HCG values were done and decreasing titer was noted. β- hCG reached normal levels at the end of five weeks, and she is on post-molar surveillance for one year while on oral combined contraceptives.

Discussion

Klippel-Trenaunay syndrome is a congenital vascular disorder with a low frequency (<1:10.000)[1]. In-about 1% of cases of KTS, a genetic pattern has been described, but the gene has not yet been identified[2]. In some cases, KTS is associated with the presence of hemangiomas in the patient's family. KTS is characterized by capillary malformations, cutaneous vascular nevi, bony or softtissue hypertrophy, and abnormal deep or superficial veins. At least two out of three main symptoms (Portwinestains, varicosity, and hypertrophy of soft tissues and bones) must be present for the diagnosis KTS. The morbidity of the disease is associated with vascular anomalies[3,4]. The first case was described by Maurice Klippel and Paul Trenaunay in 1900[7]. Later, Frederick Parkes Weber reported other cases with the same clinical findings associated with deep arteriorvenous fistula. Klippel-Trenaunay syndrome is a pure low flow condition, while Parkes Weber syndrome is characterized by significant arteriovenous fistulas[8].The etiology of the syndrome is unknown. Several theories have been proposed including abnormalities of the sympathic nerve system resulting in dilatation of the arteriovenous anastomosis or obstruction of the deep veins and persistence of fetal microscopic small arteriovenous anastomoses[9].

KTS is extremely rare in pregnant women and nearly twenty cases have been described. It puts a pregnant woman at increased obstetric risk and can increase complications, mainly thromboembolic and haemorrhagic events[10]. KTS is a high risk obstetric situation and we have to be aware of maternal and fetal complications[11]. The normal physiologic changes of pregnancy, such as increased venous pressure, leg edema, venous stasis, and cardiac output, exac-

erbate the problems of this syndrome and increase the risk of adverse events during pregnancy such as thromboembolism and haemorrhage[12]. Colour Doppler flow can display eventual uterine affectation, possibly complicating caesarean section. Coagulopathy is the most frequently reported complication during pregnancy in women with KTS, including deep venous thrombosis and other tromboembolic problems both during and after delivery. It is considered that the risk is 10 times higher than in the normal population. In spite of this, there are no prospective trials on these of anticoagulants and during pregnancy in this syndrome.If the patient had a thromboembolic event in the past, the use of anticoagulant agents is indicated[13]. Different reports on prenatal diagnosis of KTS in fetuses have been published[14]. The fetus should be checked for prenatal diagnosis of limb hypertrophy or multiloculated cystic lesions, which can be easily performed by colour Doppler flow[15]. Sometimes this syndrome has been related to intrauterine growth restriction[16]. A gynaecological examination should be made before choosing the best way of delivering in order to identifiy the presence of varicose veins in uterine cervix or vaginal wall, which could contraindicate vaginal delivery. Except for this, the delivery is chosen according to obstetric indications. Cesarean section can lead to further complications due to the existence of varicose uterine or abdominal wall viens[17]. Epidural anaesthetic is the best option during labor. The presence of neuraxial vascular anomalies and coagulopathy can increase the risk of epidural hematoma[18,19]. The management of these patients needs a multidisciplinary approach between gynecologist, anesthesiologist, hematologist and vascular surgeon.

Twin pregnancies with one normal fetus and a co-existing molar pregnancy (complete or partial) have the danger of complications for both the mother and the fetus. For the mother, in addition to haemorrhagic complications, there are increased risks of medical problems such as hyperemesis, preeclampsia, thyrotoxicosis and trophoblastic emboli. The risk of persistent trophoblastic disease (PTD) in twin molar pregnancy is more than a single complete mole, and is also increased in partial moles with a diploid fetus, compared to triploidy[20]. Preeclampsia has been reported in 34% of these pregnancies[21].

There are case reports where patients required hysterotomies and hysterectomies for severe bleeding[5,6]. The risks to the fetus when pregnancy continues in these cases include abortion, intrauterine foetal demise, pre-term labour and foetal growth restrictions. Live births in these pregnancies vary from 16.56%[22]. Another rare problem that has been reported is the molar placenta previa developing placental abscesses[23]. It has been observed that there is a tendencyfor these molar tissues to be in the lower segment of the uterus, closer to the os, thereby resulting in bleeding (on and off)[24]. Hysterectomies have recently been reported for molar placenta previa accreta, and also for spontaneous fundal rupture of the uterus in the second trimester[25,26]. The risk of PTD in CHM is said to be as high as 16.50%[22]. The risk of a partial mole developing PTD is 14-33%[27]. Stellar et al. noted a higher risk of developing PTD in cases with twin molar pregnancies with a co existent fetus, when compared to singleton molar pregnancies. PTD is not only more common, but it requires multiple cycles of combination chemotherapy[28]. Another interesting observation made in some studies is that PTD was more often seen in those twin molar pregnancies with maternal complications such as preeclampsia, hyperemesis, etc.[29].

Prenatal diagnosis by amniocentesis has been advocated to rule out triploidy or any other genetic abnormalities, before advising the continuation of a pregnancy. Chorionic villi sampling for karyotype is not recommended, as it may vary from that of the fetus because of confined placental mosaicism. Most often, the triploid fetus co-existing with a partial mole tends to die, while a fetus with a complete mole tends to survive[30-32].

It has been reported that very high β hCG (> 106 mIU/mL) and the presence of medical complications portend a poor outcome, and may serve as an indication for the termination of pregnancy because they

are suggestive of aggressive trophoblastic growth [32-35]. Fortunately, our patient did not develop any medical complications.

Regarding surgical evacuation for gestations less than 18 weeks, several osmotic dilators should be placed on the day before the procedure[36]. Misoprostol is an attractive agent because it is inexpensive, stable at room temperature, and easily applied either orally or vaginally. At 14 to 16 weeks' gestation, per os misoprostol (600 ìg) two to four hours prior to D&C can provide dilatation and softening for vacuum aspiration with a 14 mm curette or further dilatation[37]. If misoprostol is administered vaginally more than four hours before the procedure, there is potential for unexpected delivery (greater than with osmotic dilators). D&C can be performed safely under local anaesthesia (paracervical block) with intravenous conscious sedation and analgesia. The most important requirements for safe D&C are the specialty training, skill, and experience of the surgeon[38]. If multiple or serial osmotic dilators are used it is usually unnecessary to dilate the cervix further. Up to 17 weeks' gestation, the uterus can generally be evacuated with a number 16 curette or extraction forceps. After 17 weeks' gestation, the amniotic fluid should be carefully and slowly emptied with a suction curette following which the POC should be removed with extraction forceps[36].

It is suggested that during forceps extraction the physician keep one hand on the fundus as a splint to reduce the risk of perforation. The procedure may also be performed under ultrasound guidance in an attempt to minimize the incidence of perforation. The fetal tissue should not be removed forcibly through the cervix, as bone spicules may lacerate the cervix. Crushing and rotating techniques lessen cervical trauma[38]. After forceps extraction, the uterine cavity should be gently explored with a large curette to ensure complete evacuation. The products of conception need to be examined for completeness.

Zhang P et al. [39] from University of California San Diego reported partial molar pregnancy with dead fetus in utero at 26 weeks. Zahida P et al. in Pakistan[40], reported G4P1+2 lady with partial hydatidiform mole along with a live baby managed conservatively 18 weeks onwards and delivered successfully at term. Bruchim et al.[41] reported a case of a lady with partial mole at 41 weeks of gestation and another at 26 weeks with complete hydatidiform mole along with twin fetuses. Tamrakar et al. (2011)[42] reported a case of preterm gestation along with partial hydatidiform mole and a live fetus. Such patients have risk of developing persistent gestational trophoblastic disease. Patient with molar pregnancy can develop choriocarcinoma and mortality has been reported by Seckl et al.[10]. However partial hydatidiform mole rarely requires chemotherapy. Several factors influence the outcome of the fetus in partial molar pregnancy most important being karyotype of the fetus. The problems in the management of molar pregnancy and a live fetus involve the risks of fetal abnormality, malignant trophoblastic change, and severe maternal complications such as preeclampsia, thyrotoxicosis, heavy bleeding, pregnancy failure, and preterm birth. Termination of pregnancy might be required due to these complications. Amniocentesis should be done for karyotyping. Prognosis of partial mole is usually better than the complete mole as only few cases of partial moles progress to persistent trophoblastic disease. However, the nature and the risks of diploid partial moles are not well established and they seem to be a distinct clinical entity.

In absence of gross fetal abnormalities on sonography, some authors recommend to continue the pregnancy as long as maternal complications are absent or controllable. Complete evaluation of the placental tissue is important even in cases with normal fetal outcome as focal molar changes, which might be unsuspected during antenatal period, may affect the future obstetrical outcome[43].

Conclusions

Klippel-Trenaunay Syndrome (KTS) is a rare syndrome of congenital vascular malformations. Pregnancies with a hydatidiform mole and a live fetus are extremely rare. This is the first case to report this association. When detected early in pregnancy, the

management approach is generally to offer termination in view of the anticipated complications. Nevertheless, it seems reasonable to opt for conservative management with the consent of the patient, after thorough counselling. However, one must be prepared for termination in the event of maternal complications like severe preeclampsia or bleeding. The most important requirements for safe D&C are the specialty training, skill, and experience of the surgeon. Induction of abortion is safe for VM with a fetus with low risk of theoretical trophoblastic embolization especially with low fetus over the cervix or advanced pregnancy with partial mole or twin with molar pregnancy.

Conflict of interest / Ethical Standards

Funding: This study was not funded .

Conflict of Interest: Authors have nothing to declare.

Ethical approval: All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent: Informed consent was obtained from the patient included in the study.

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