A hydatidiform mole coexisting with a normal live fetus in a twin pregnancy is an extremely rare phenomenon, with a worldwide incidence ranging from one in 22,000 to one in 100,000 pregnancies. This condition presents a significant management challenge for physicians due to its associated complications, including heavy vaginal bleeding, severe pre-eclampsia, intrauterine fetal death or growth restriction, miscarriage, preterm birth and hyperthyroidism, as well as ovarian cyst rupture or torsion in cases with theca lutein cysts. Twin molar pregnancies may also have an increased risk of persistent trophoblastic disease compared to a single molar pregnancy.

Case Report

A 32-year-old gravida 3, para 2 woman was admitted to the Emergency Department of the Sultan Qaboos University Hospital (SQUH), Muscat, Oman, in March 2012 at 13 gestational weeks with lower abdominal pain and vaginal spotting. Upon examination, she was haemodynamically stable and not in distress. Abdominal palpation indicated mild tenderness and a uterine size corresponding to 16 gestational weeks. An ultrasound examination revealed a twin pregnancy, including one normal fetus with the placenta covering the cervical opening and one molar pregnancy [Figure 1A]. Both ovaries were enlarged with multiple theca lutein cysts; the right ovary measured 12.2 x 8.1 x 6.9 cm while the left measured 13.3 x 7.3 x 10.6 cm [Figure 1B]. The patient’s serum β-human chorionic gonadotropin (hCG) level was 1,386,570 IU/L. Chest X-ray findings were normal as were the results of the thyroid function tests and coagulation profile.

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were elicited and the couple was informed that a review of the literature indicated a 25% chance of a live birth and a 40% chance of early fetal loss. A multidisciplinary team including a consultant haematologist, an anaesthetist, an interventional radiologist and several obstetricians were involved in the decision-making process. After consideration, the couple opted to continue the pregnancy with close follow-up and the patient was discharged after three days.

Despite close follow-up, the patient was readmitted at 17 gestational weeks with vaginal bleeding and tachycardia. A significant drop in haemoglobin from 10 g/dL to 7 g/dL was also noted, requiring a transfusion of two units of packed red blood cells. At this time, the uterine size corresponded to 28 gestational weeks. The couple were advised that the maternal risks had now increased significantly and they agreed to have the pregnancy terminated. Bilateral uterine artery balloon catheters were placed prophylactically and inflated to control the intraoperative blood loss; this provision would also enable the postoperative embolisation of the uterine arteries if required.

A hysterotomy was performed under general anaesthesia via a lower segment transverse incision. Molar tissue weighing 1,040 g was evacuated, along with a fetus weighing 105 g and a normal placenta of 65 g [Figure 2]. The ovaries were enlarged with bilateral theca lutein cysts. The intraoperative blood loss was 800 mL. The uterine artery balloon catheters were removed six hours after the operation as there was no active bleeding and embolisation was not deemed necessary. The postoperative recovery period was uneventful. Histopathology results confirmed a complete hydatidiform mole with a coexisting fetus and normal placenta [Figure 3]. Three months after the surgery, the patient’s serum β-hCG level had returned to normal without any cytotoxic therapy. The enlarged ovaries with multiple theca lutein cysts took six months to return to a normal size. There was no evidence of any persistent trophoblastic disease one year later and the serum β-hCG level remained normal for 18 months following the surgery. At the time of writing, the patient was planning her next pregnancy.

Discussion

Complete hydatidiform moles form part of the tumour spectrum of gestational trophoblastic disease. Molar pregnancies are subdivided into either complete or partial types based on genetic and histological features. A complete hydatidiform mole consists of oedematous villous tissue and trophoblastic hyperplasia with no fetal tissue. While a complete mole is diploid, its chromosomes are entirely of paternal origin, following the duplication of a single sperm fertilising an empty ovum. However, the majority of partial moles are triploid in origin and contain two sets of paternal and one set of maternal haploid genes. This occurs following the fertilisation of a normal ovum by two sperm cells. In a partial mole, a fetus or fetal red blood...
Twin Pregnancy with a Complete Hydatidiform Mole and a Coexisting Live Fetus

Rare entity

past decade, clinicians have supported the option of conservation, providing that the patient remains under strict hospital-based observation and follow-up.8,9 With the current medical facilities available, most expected complications can be diagnosed early and managed appropriately. Although the risk of persistent trophoblastic disease cannot be excluded in cases of conservative management, this risk does not seem to increase with gestational age.10,11 Conservative management in a tertiary care environment is indicated if a multi-disciplinary team is present and able to quickly respond in the event of an emergency. Close surveillance of the patient is also recommended to detect early signs of maternal and fetal complications. Termination of the pregnancy should be considered only if the patient develops severe pre-eclampsia, thyrotoxicosis or significant vaginal bleeding, or if there is evidence of trophoblastic embolisation.12

Measures to minimise blood loss during surgery for complete hydatidiform twin molar pregnancies with live co-twins are not described in the literature, although torrential haemorrhage is an expected complication. As such, the authors of this report strongly recommend preoperative placement of bilateral uterine artery balloon catheters for patients at centres with interventional radiology facilities. This will assist in decreasing the intra- and postoperative blood loss and minimise the need for blood and blood product transfusions.

Conclusion

In twin pregnancies with one complete hydatidiform mole, the likelihood of delivering a healthy viable fetus is low. This type of pregnancy creates a

Figure 3A & B: Haematoxylin and eosin stains of the molar tissue showing (A) normal trophoblasts (arrow) at x10 magnification and (B) avascular oedematous villi (arrow) in a pregnant patient with a complete hydatidiform mole and a coexisting live fetus.
significant management dilemma for the treating physician, especially if the pregnancy was planned. Comprehensive counselling, close follow-up and timely intervention are imperative during the conservative management of this rare clinical entity.

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