CASE REPORT

The forgotten child—a case of heterotopic, intra-abdominal and intrauterine pregnancy carried to term

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Heterotopic pregnancies are estimated to be less frequent than one in 30 000 if no assisted reproduction technologies are performed. Here we report a case which occurred in Tanzania. An abdominal pregnancy at term was first misdiagnosed as an ovarian tumour and diagnosed on the first post-partum day of the intrauterine fetus, which was delivered spontaneously. The abdominal pregnancy was then treated by laparotomy and removal of the placenta. The fetus was alive and healthy. The follow-up of the twins was normal.

Key words: abdominal pregnancy/ectopic pregnancy/heterotopic pregnancy

Introduction

Heterotopic pregnancies in natural cycles are a very rare event. The first case was reported in 1708 as an autopsy finding (Bright and Gaupp, 1990) and this condition is estimated to be less frequent than one in 30 000 pregnancies (Reece et al., 1983).

We report on a heterotopic pregnancy, which was diagnosed in Tanzania in the third trimester and both fetuses, the ectopic and the orthotopic, were delivered at term.

Case report

The gravida 9, para 9 patient was 39 years old. Her last pregnancy was a twin pregnancy. At 38 weeks of gestation she was referred in labour to the Muhimbili Medical Centre, Tanzania, East Africa. Besides labour pains, her main complaint was a swelling in the right upper abdomen, reaching the right hypochondrium. The swelling was diagnosed by the referring doctor as a hepatomegaly or an ovarian tumour. This swelling had been noticed by the patient 4 months prior to admission. Initially she described it as starting in the right iliac fossa, increasing rather slowly in size. Compared to her previous twin pregnancy, she felt excessive fetal movements and different abdominal distension.

Two hours prior to admission, she had spontaneous rupture of the membranes. The liquor was clear, there was no vaginal bleeding. The diagnosis of a full-term singleton pregnancy with an ovarian tumour was made, and a spontaneous vaginal delivery of a healthy baby boy weighing 3000 g, Apgar 9, 10, and 10, occurred. The early postnatal period was normal, but the tumour persisted and was now found to extend from the right iliac fossa to the left iliac fossa. It was firm, non-tender and mobile. Now bilateral ovarian tumours were suspected, and the patient was not to be discharged until after surgery.

The first day post partum, the patient reported fetal movements and continuous lower abdominal pain, dull in character, non-radiating and not relieved by analgesics. The first time an abdominal ultrasound was performed, it revealed a fetus in breech in the pouch of Douglas behind the uterus, with a biparietal diameter equivalent to 38 1/2 weeks of gestation. The uterus was empty and the adnexa were normal.

On the fourth post-partum day a laparotomy was performed, which confirmed the diagnosis of an intra-abdominal pregnancy. A fetus in transverse lie was found, in its intact amniotic sac posterior to the uterus, in the pouch of Douglas. The head was in the right iliac fossa behind the placenta, which was attached between the anterior and posterior leaves of the right broad ligament. The main blood supply to the placenta was identified as arising from the right ovarian vessels. The right Fallopian tube and right ovary could not be identified, the left Fallopian tube and ovary were normal.

After opening the amniotic cavity a healthy baby boy was delivered. The placenta was removed from the abdomen in total by unilateral adnexectomy, using successive clamps and interrupted chromic catgut no. 1 sutures, which were applied below the clamps to tie off the blood vessels.

The postoperative period was uneventful, and the patient was discharged on the sixth postoperative day with both babies, which were in good health and on breast milk. The follow-up showed no abnormalities.

Discussion

In a twin heterotopic pregnancy, the chance that both children are carried to term and survive the neonatal period is very low: Reece et al. (Reece et al., 1983) identified 13 cases in 589 reports from the world literature where this condition is described.

The diagnosis of an abdominal pregnancy is even more complicated than that of a heterotopic pregnancy with one embryo located in the Fallopian tube. Disappointing results were shown in a study (Martín et al., 1988), who reported on
15 cases of abdominal pregnancies. This is confirmed by others (Ali et al., 1981; Brown et al., 1984). The most important problem is that the sonographer has to be aware of the possibility of an advanced abdominal pregnancy. One has to identify the uterus and the fetal head, which is outside the uterine cavity. Sonography is also helpful in that, from a lateral projection, the fetal skull may overlie the maternal spine. Fetal malpresentation as transverse lie, the identification of an oligohydramnion (Cartwright et al., 1986; Moessinger, 1986) and malformations (Tan et al., 1971) should, especially when occurring in combination, arouse suspicion. Hysterography was found to be the most helpful test in making the diagnosis of an abdominal pregnancy (Martin et al., 1988). Also helpful seem to be a radiograph of the maternal abdomen and pelvic area, especially from a lateral view, which can demonstrate small fetal parts in the maternal abdomen, in an abnormal relationship to the expected normal position.

Other diagnostic tools are the lack of myometrial response to oxytocin stimulation (Chessin and Zussman, 1954). Because amniotic fluid may be transferred in larger amounts to the maternal circulation, the measurement of an abnormal high maternal serum α-fetoprotein should also raise the differential diagnosis of an abdominal pregnancy (Tromans et al., 1984).

The condition described here, i.e. a surviving ectopic tubal pregnancy, is rare but also described in the literature for isolated ectopic pregnancies (Kranzfelder et al., 1988), and also for abdominal pregnancies even after diagnosis is made in the second trimester (Bassil et al., 1991). However, it is not higher than 10.6% (Vesicka, 1956) when reviewing 435 cases. This case shows an additional risk factor for heterotopic pregnancies, apart from tubal damage and assisted reproductive treatment, which is multiparity. In addition, as the incidence of twinning is higher in Africa than in Europe, Asia and America, the incidence of heterotopic pregnancy in Africa might also be higher (Bulmer, 1960), though less frequently reported. The highest rate of ectopic pregnancies in non-Caucasian women were reported (Maymon et al., 1992).

Two cases in Kitui district hospital were reported (Madhany, 1977) in Kenya and one case was reported (Tengio et al., 1983) in Bagamoyo District Hospital. Two cases have occurred in the new maternity block of the hospital in Dar es Salaam (unpublished data). An incidence of one in 3259 deliveries over a 2 year period was found at the Muhimbili Medical Centre (Mbura and Mgaya, 1986). The incidence of abdominal pregnancy as reviewed in the world literature is as high as one in 8000, with a perinatal and maternal mortality of 75–95% and 2–18% respectively (Delke et al., 1982). Only 1.6% of all ectopic pregnancies are found to be abdominal (Golz et al., 1984). The incidence of congenital malformations due to the abdominal location of the pregnancy is estimated to be 30–90% (Tan et al., 1971).

In an advanced heterotopic pregnancy such as this one, the extraterine pregnancy is usually abdominal, as described (Bassil et al., 1991), or ovarian. As such, the great difficulty lies in the management of the co-existent extrauterine pregnancy.

The diagnosis would have been easy if an ultrasound examination were a standard procedure, when patients enter the labour ward, as is usual in Europe. It would have been easy, retrospectively, if the idea had occurred to someone that a heterotopic pregnancy could exist. At no time that the patient in this case was seen, within the first 24 h of hospitalization, was the diagnosis of heterotopic pregnancy suspected. In fact, it was the patient's perception of fetal movements that guided us to the diagnosis, which was confirmed by sonography. If the diagnosis had been made before delivery of the intrauterine pregnancy, the extraterine pregnancy would have been delivered by laparotomy and the intrauterine one by Caesarean section.

The other significant problem at operation is whether or not to remove the placenta. Interference with it may lead to uncontrollable haemorrhage and if it is left in situ, though it is usually the procedure of choice, the morbidity from abscess formation is high (Babic et al., 1983). Removal of the placenta should be undertaken if it is safe, depending upon the accessibility of ligation of the maternal vessels supplying the placenta (Foster and Moore, 1967; Golz et al., 1984; White, 1989). This reduces hospital stay and maternal morbidity (Foster and Moore, 1967; Mbura, 1983), as was the case in this patient, who was discharged home on the sixth postoperative day, healthy.

A non-surgical approach (Hreshchyshyn et al., 1965), who reported on methotrexate treatment in a patient with abdominal pregnancy after removal of a dead fetus in the fifth month of pregnancy. The placenta could not be removed surgically, and after four methotrexate administrations (0.2 mg/kg body weight) human chorionic gonadotrophin titres went down within 35 days. After that, eight more administrations of methotrexate at the same dose were given and 3 months after the first laparotomy the placenta was removed surgically.

The case presented here shows a typical problem of medicine: the failure to consider alternatives after a primary diagnosis has been made. In this case, the diagnosis was ovarian tumour in a pregnancy at term. The possibility of a heterotopic pregnancy was not considered until the patient herself mentioned fetal movements after spontaneous vaginal singleton delivery.

References


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