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Case report

Cervical pregnancy ending in a live vaginal birth

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A case of cervical pregnancy followed by cataclysmic haemorrhage estimated at 8500 ml and necessitating total abdominal hysterectomy and replacement of 18 units of blood is reported. This pregnancy remained undiagnosed until 28 wk and is to our knowledge the first case described in the English literature in which the pregnancy ended in a live vaginal birth. Both mother and baby are now well.

cataclysmic haemorrhage; first case in English literature: 28 wk gestation

Introduction

Cervical pregnancy is extremely uncommon and rarely progresses beyond 20 wk gestation (Beisher, 1964; Ratten, 1983). Diagnostic ultrasound has been widely used in obstetrics in recent years and can readily detect cervical pregnancy (Kobayashi et al., 1969; Raskin, 1978; Chow et al., 1979; Gitstein et al., 1979; Pridmore et al., 1980; Giorgio et al., 1980; Szeja et al., 1980; Williams et al., 1982; Zarabi et al., 1983), allowing conservative measures to be instituted before catastrophic haemorrhage ensues. Baptisti (1953) stated: 'The great majority of obstetricians will never see a cervical pregnancy. The minority who do happen to encounter this complication will probably wish they had not'. We report a case which was not diagnosed by ultrasound and in which the pregnancy continued until 28 wk of gestation.

Case report

Mrs A.P., a 43-yr-old Greek lady, para 1 + 3, presented with premature labour at 28 wk of pregnancy. She was transferred to the Whittington Hospital from the City of London Maternity Hospital, where no neonatal facilities were available.

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TABLE I

Pallman's and McElin's clinical signs for the diagnosis of cervical pregnancy

1. Uterine bleeding after amenorrhoea and without cramping pain.
 2. A softened and disproportionately enlarged cervix equal to, or larger than, the corporeal portion of the uterus, and an hour-glass shaped uterus.
 3. Products of conception entirely confined within and firmly attached to the endocervix.
 4. A snug internal os.
 5. A partly open external os.
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Her past obstetric history consisted of three first-trimester spontaneous abortions between 1969 and 1974 requiring curettage, and in 1975 she was delivered of a male infant weighing 3.2 kg by elective lower-segment caesarean section at term because of her age and poor obstetric history.

In this pregnancy she presented to the referring hospital at 11 wk gestation by dates, with a threatened abortion. The uterus corresponded in size to 14 wk gestation. An ultrasound scan showed a single fetus with a crown-rump length of 45 mm compatible with 11–12 wk gestation and no abnormality was seen. She was advised to rest but continued to have a slight blood-stained vaginal discharge. An ultrasound scan at 19 wk gestation agreed with the dates and was reported to show a fundal placenta; again no abnormality was noted. An offer of amniocentesis because of her age was declined.

At 24 wk gestation she presented as an emergency with a small amount of vaginal bleeding. The cervix was dilated to 2 cm. An incompetent cervix was diagnosed and cervical cerclage was performed by the McDonald technique using monofilament nylon.

She had a further emergency admission at 27 wk gestation with spontaneous rupture of membranes, without uterine contractions. The cervical suture was found to be loose and was removed. She was kept at rest in bed.

A week later, on the day of her transfer to the Whittington, she complained of low backache. An ultrasound scan the same day showed a biparietal diameter compatible with 28 wk gestation and again the placenta was reported in the fundus.

Two days after admission she complained of low backache and tenesmus. On vaginal examination the cervix was found to be fully dilated. After a prolonged second stage, with a satisfactory continuous fetal heart tracing, she was delivered of

TABLE II

Rubin's pathologic criteria for the diagnosis of cervical pregnancy

1. Cervical glands must be present opposite the placental attachment.
 2. The attachment of the placenta to the cervix must be intimate.
 3. The whole or a portion of the placenta must be situated either below the entrance of the uterine vessels or below the peritoneal reflexion of the anterior and posterior surfaces of the uterus.
 4. Fetal elements must not be present in the corpus uteri.
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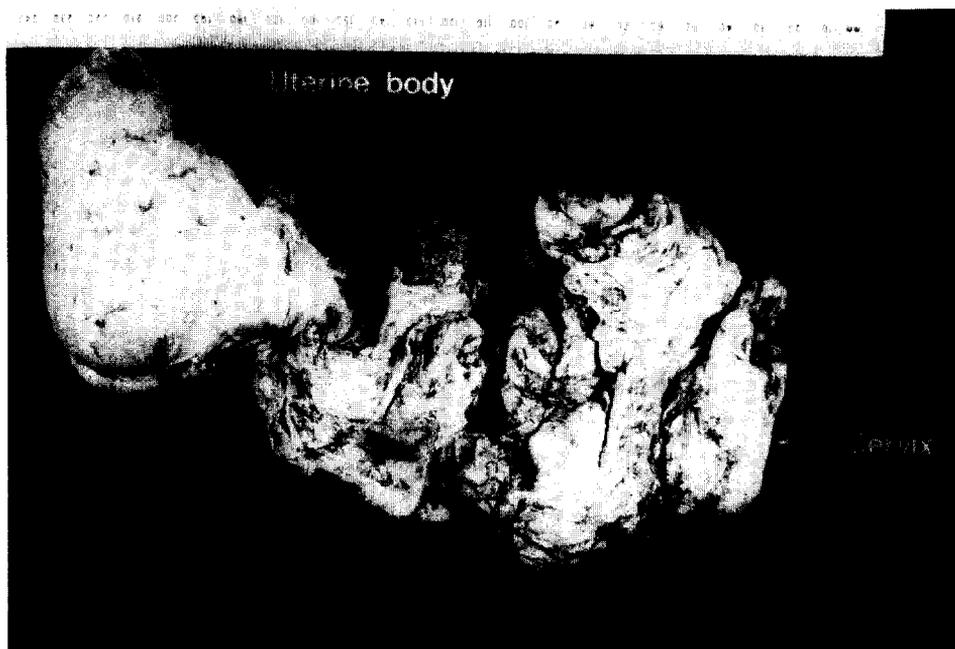


Fig. 1. The surgical specimen shows the small uterine body situated superior to the ballooned and ragged cervix. This case fulfils all of Paalman's and McElin's (1959) clinical signs (Table I) and Rubin's (1911) pathologic criteria (Table II) for cervical pregnancy.

a female infant with Kjelland's forceps. The baby had an Apgar score of 2 at 1 min, weight 1.1 kg, was intubated immediately and transferred to the Special Care Baby Unit.

The third stage was complicated by heavy bleeding and difficulty in delivering the placenta, requiring manual removal under general anaesthetic. The placenta was found to be adherent posteriorly, but was completely removed. A rapid 2-litre post-partum haemorrhage followed, which was not controlled by uterine massage, compression and oxytocics. The blood pressure dropped from 130/80 mmHg to 70/50 mmHg. Blood transfusion was started immediately and an emergency laparotomy was performed. At laparotomy, there was no blood in the peritoneal cavity.

A ballooned, distended, thin structure, continuous with the lower end of the uterus, arose from and filled the pelvis. The small uterine body was clearly seen as a separate discreet body on top of this mass (Fig. 1). The distended cervix was thin with ragged edges, friable and bled easily on gentle manipulation. A difficult hysterectomy was carried out in which sponge holding forceps were applied vaginally to the cervix as markers to facilitate the difficult excision of the thin friable haemorrhagic cervix.

The total estimated blood loss was 8500 ml. This was replaced with 18 units of blood administered through filters and blood warmers.

In addition, crystalloid and colloid solutions, 2.5 litres of plasma protein fraction,

6 units of fresh frozen plasma and 1 unit of dried plasma were administered. Her blood pressure at the end of the operation rose to 100/70 mmHg.

Macroscopy of hysterectomy specimen

The body of the uterus was $6.8 \times 9.2 \times 5.5$ cm with necrotic tissue-filled cervix $16.2 \times 13.5 \times 2$ cm thick with ragged lining. Histology showed an extensively infarcted placenta with irregular penetration of endocervical muscle. The baby was resuscitated in the Special Care Unit and treated for streptococcal chest and eye infections. Jaundice was treated with phototherapy. The baby was discharged home when 86 days old, weighing 2.44 kg.

Both mother and baby are now alive and well.

Discussion

The incidence of cervical pregnancy is rising (Parente et al., 1983) and varies from 1 in 1000 pregnancies in Japan (Shinagwa and Nagayama, 1969) to 1 in 95 000 in other countries (Paalman et al., 1959).

This case illustrates the life-threatening haemorrhage which follows delivery of late cervical pregnancy (Rubin, 1911; Studdiford, 1945; Schneider et al., 1957; Paalman et al., 1959; Sherwin et al., 1960; Jauchler et al., 1970; Parente et al., 1983). Haemorrhage is the result of trophoblastic invasion into the non-retractile cervical tissue. Maternal vessels are exposed after separation of the placenta and, as they are not surrounded by contractile fibres, the natural defences against post-partum haemorrhage are absent. The entire placental blood flow of some 450 ml per min confronts the obstetrician with a dramatic and frightening spectacle. In retrospect, the diagnosis should have been made by ultrasonography at the referring hospital. However, the unwary ultrasonographer who fails to notice the uterine fundus situated superiorly to the products of conception will easily overlook this condition (Kobayashi et al., 1969; Raskin, 1978; Chow et al., 1979). The characteristic features consist of a bulky fundus superior to an enlarged cervix containing the gestational sac; the two combining to give a dumb-bell or hour-glass shape to the uterus (Gitstein et al., 1979; Pridmore et al., 1980; Giorgio et al., 1980; Szega et al., 1980; Williams et al., 1982; Zarabi et al., 1983). Planned evacuation soon after diagnosis in the first half of pregnancy can avert massive haemorrhage and the need for hysterectomy (Gitstein et al., 1979; Giorgio et al., 1980; Williams et al., 1982; Kuppuswami et al., 1983; Bernstein et al., 1981).

In these cases it has been suggested that the placenta should be left undisturbed to await natural absorption (Morton, 1949).

This case was unusual in that the pregnancy continued until 28 wk and ended in a live vaginal delivery. The only other recorded cervical pregnancies resulting in live births ended in caesarean hysterectomy (Chiner and Ballester, 1948; Pisarski, 1960; Mitrani, 1973). Abortion usually takes place before 20 wk. Management in these cases includes evacuation of the uterus followed by specific measures to arrest haemorrhage. These include cervical packing, compression with catheter balloon (Kuppuswami et al., 1983; Patchell, 1984), compression with tampon (Gitstein, 1979), cervical circumsture (Giorgio et al., 1980; Woodford Scott et al., 1978; Bernstein et al., 1981), ligation of cervical branch of uterine arteries (Kuppuswami et

al., 1983; Ratten, 1983) and bilateral iliac artery ligation (Nelson, 1979). After 20 wk hysterectomy is almost mandatory, as in this case (Studdiford, 1945; Baptiststi, 1953; Ratten, 1983).

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