

# A case of a cornual heterotopic pregnancy laparoscopically treated with the endoloop technique

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**Abstract** Heterotopic pregnancy is defined as the coexistence of an intrauterine and an ectopic pregnancy. The estimated incidence is one in 30,000 spontaneous pregnancies, with a tenfold increase in women who underwent assisted reproductive technologies. Diagnosis of a heterotopic pregnancy is often delayed because of the presence of the intrauterine gestational sac. Treatment of a heterotopic pregnancy should consist of termination of the ectopic pregnancy without damaging the ongoing intrauterine pregnancy. The least invasive procedure should therefore be used. We present a case of a heterotopic pregnancy consisting of a viable intrauterine pregnancy and an ectopic cornual pregnancy. Because of the viable intrauterine pregnancy, we decided to treat the cornual pregnancy laparoscopically by the endoloop technique. This technique is simple, safe, effective and nearly bloodless. It offers a good prognosis for the ongoing intrauterine pregnancy.

**Keywords** Heterotopic pregnancy · Cornual pregnancy · Laparoscopy · Endoloop technique

## Introduction

Heterotopic pregnancy is defined as the coexistence of an intrauterine and an ectopic pregnancy. It is a rare form of a twin pregnancy, with an estimated incidence of one in 30,000 spontaneous pregnancies. There is a tenfold increase

in incidence in women who underwent assisted reproductive technologies (ART) [1–3].

An interstitial or cornual pregnancy is an uncommon form of an ectopic pregnancy occurring once in every 2,500 to 5,000 pregnancies and accounting for 2–4% of all ectopic pregnancies. The interstitial part of the fallopian tube is the proximal portion of the tube that lays within the muscular wall of the uterus. Maternal mortality in case of cornual rupture is described to be 2–2.5% [2, 4].

The incidence of heterotopic pregnancy with the intrauterine pregnancy located in the cornual area is not exactly known [5].

Heterotopic pregnancies are usually diagnosed at a later gestational age than other ectopic pregnancies because of the presence of an intrauterine gestational sac. If rupture of the cornual part occurs, haemorrhage can be dramatic.

Historically, cornual pregnancies have been treated by laparotomy with hysterectomy or cornual resection. In the last decades, however, several conservative techniques have been introduced: expectant follow-up with regular sonography, local or systemic use of methotrexate, hysteroscopic removal and laparoscopic resection. In case of the coexistence of a viable intrauterine pregnancy and an interstitial pregnancy, some treatment options, such as the use of chemotherapeutic agents, cannot be considered [2, 4].

We present a case of a heterotopic cornual pregnancy in which we used the endoloop technique to extract the corneal pregnancy. To our knowledge, this is the second report on the use of this technique in the case of an interstitial heterotopic pregnancy [6].

## Case report

A 36-year-old woman became pregnant after the third cycle of in vitro fertilisation (IVF) with intracytoplasmic sperm

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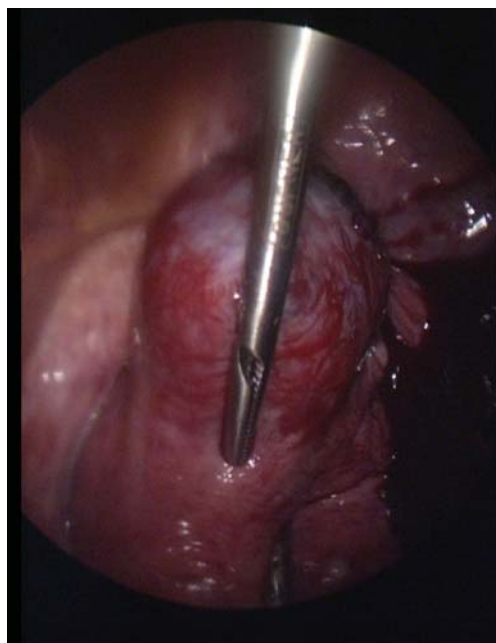
injection. The reasons for infertility were tubal pathology with a male factor. Ten years before this IVF treatment, a first diagnostic laparoscopy was performed because of chronic abdominal pain. Bilateral hydrosalpinges, extensive tubo-ovarian adhesions and perihepatic adhesions were visualised, probably as a result of an inflammatory process. At that time, a bilateral salpingostomy and adhesiolysis were performed. A few years later, a second laparoscopy was performed because of pain and infertility problems. Again, there were strong tubo-ovarian adhesions but no evident hydrosalpinges. Because she already had a bilateral salpingostomy and a problem of infertility, we decided not to perform a second adhesiolysis but to start an IVF treatment. A hysteroscopy was not performed before starting the IVF treatment.

Four weeks after intrauterine transfer of two embryos, a viable singleton intrauterine pregnancy was visualised on transvaginal ultrasound.

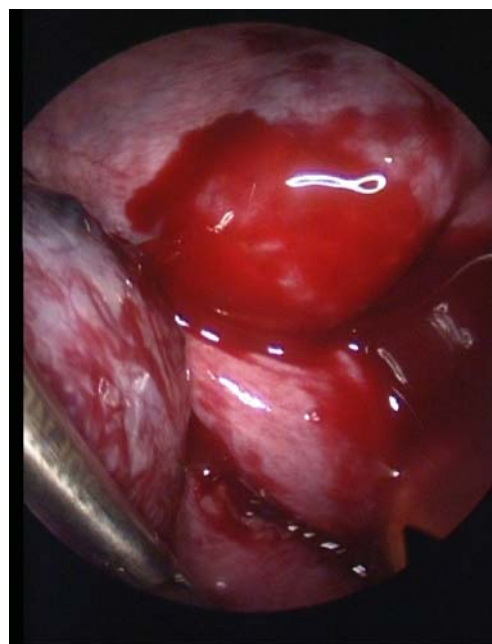
At a gestational age of 9 weeks, she was admitted to the emergency room because of severe lower abdominal pain and syncope. Clinical examination revealed signs of an acute abdomen. There was no vaginal bleeding. She was haemodynamically stable with a tension of 120/70 mmHg and a heart rate of 76. The hematocrit was 36.9%.

Ultrasonography demonstrated a viable intrauterine pregnancy with crown–rump length of 31 mm and bilateral multicystic ovaries (60×50 mm) as a result of the ovarian hyperstimulation. The presence of blood in the pelvic cavity was suspected.

A few hours after admission, we decided to perform a laparoscopy because of the increasing pain and the presence



**Fig. 1** Right cornual area, slightly pedunculated from the uterine fundus and partially ruptured at the base



**Fig. 2** Cornual pregnancy was partially ruptured at the base and constantly bleeding

of free fluid in the pelvic cavity. Because of the enlarged ovaries, there was suspicion of an ovarian torsion. A heterotopic pregnancy was not really suspected before performing the laparoscopy.

At laparoscopy, we saw a cornual pregnancy at the right side. The cornua, which was slightly pedunculated from the uterine fundus, was partially ruptured at the base and constantly bleeding. Approximately 500 ml of blood was present in the abdomen (Figs. 1 and 2).

Because of the presence of a viable intrauterine pregnancy, we decided to treat the cornual pregnancy conservatively. A first endoloop (Ethicon, Johnson & Johnson Medical NV, Dilbeek, Belgium) was loosely tightened around the elevated cornual area (Fig. 3). Using unipolar coagulation, an incision was made in the myometrium over the cornua. While fastening this first loop, the conceptus was gradually removed with a grasper and a suction tube. After removal of all trophoblastic tissue and coagulation of the underlying myometrium, a second endoloop was consecutively fastened around the tubal stump to secure hemostasis (Fig. 4). The amputated cornua was removed from the abdomen through an endobag. Pathological examination showed tubal tissue and placental villi.

The postoperative course was uncomplicated, and in the next few weeks the intrauterine pregnancy progressed uneventfully.

At a gestational age of 12 weeks, however, she was readmitted because of increasing pain in the right ovarian fossa and vomiting. Transvaginal ultrasound showed a viable intrauterine pregnancy. At the right cornual area, we saw a hyperechoic mass. A repeat laparoscopy was



**Fig. 3** A first endoloop was loosely tightened around the bleeding cornual area

performed because of suspicion of residual trophoblastic tissue in the right cornua. On laparoscopy, the stump of the right cornua was still slightly swollen with fibrin tissue covering it, but without evidence of residual trophoblastic tissue or bleeding. A biopsy was taken of the right cornual stump, and histological examination showed fibrine, decidual tissue and mesothelial hyperplasia, but no trophoblastic tissue. The postoperative course was again uncomplicated.

Afterwards, the pregnancy continued well and a caesarean section was planned at 38 weeks. However, at a gestational age of 28 weeks, she was admitted to the delivery room because of preterm premature rupture of the membranes. As contractions started shortly after, tocolysis (Ritodrine®) and lung ripening (Betamethasone®) were initiated. Despite tocolysis, labour progressed rapidly, and as there were no signs of uterine rupture, we decided to accept a vaginal delivery. She delivered a healthy premature daughter of 1,200 g with Apgar scores of 10 and 10 after 1 and 5 min. Because of light respiratory problems, she was treated with nasal continuous positive airway pressure for 5 days. The child could leave the neonatal care unit 7 weeks later. There were no signs of neurological, ophthalmological or respiratory sequelae.

## Discussion

The incidence of heterotopic pregnancy has steadily increased since the development of ART and is estimated at 0.3% with the use of ART. Despite the growing incidence, heterotopic pregnancies still remain a diagnostic

and therapeutic challenge. Gynaecologists should be vigilant in any case of a pregnancy after ART with persistent bleeding and abdominal pain. Repeated oriented pelvic ultrasound examinations are therefore recommended [3, 7, 8].

When in IVF procedures multiple embryos are transferred, the multiple gestation rates, including those of heterotopic pregnancies, increase. The factors commonly associated with interstitial pregnancy include pelvic inflammatory disease, previous pelvic surgery, pelvic tumors, congenital uterine anomalies and the use of ART [3–5, 7, 9]. In our case, no uterine anomalies were seen at prior laparoscopy. A hysteroscopy was not performed.

A predisposing factor for heterotopic pregnancy is multiple embryo transfer. Several reports describe the occurrence of triplet and quadruplet heterotopic pregnancies after transfer of multiple embryos. Current guidelines all advise single embryo transfer in young women to reduce the multiple gestation rate and thereby the heterotopic pregnancy rate [10–12].

The diagnosis of a heterotopic pregnancy is difficult and symptoms are often misleading. The most common clinical signs are abdominal pain, presence of an adnexal mass and peritoneal irritation. The majority of symptoms occur as a result of bleeding from the ectopic pregnancy. Haemorrhagic shock may occur in case of tubal rupture. Abdominal pain without vaginal bleeding is often reported, and the absence of vaginal bleeding is suggestive for the diagnosis of a heterotopic pregnancy rather than an ectopic pregnancy alone [1, 3, 13].



**Fig. 4** After removal of all trophoblastic tissue and coagulation of the myometrium, a second endoloop was fastened around the tubal stump to secure hemostasis

Diagnosis is often delayed, with only less than half of cases diagnosed before operative intervention [3, 5, 7].

Transvaginal ultrasound is an important aid in the diagnosis of interstitial heterotopic pregnancy. Careful sonographic assessment of the whole pelvis is critical. Several ultrasonographic findings have been used to diagnose a cornually located pregnancy: a whole bridge of myometrium separating the twin gestations and a chorionic sac seen separately and more than 1 cm from the most lateral edge of the uterine cavity, with a thin myometrial layer surrounding the chorionic sac [5, 13].

The simultaneous presence of an intrauterine pregnancy may cause difficulties in the interpretation of ultrasound images. Furthermore, the diagnosis of an ectopic pregnancy may be difficult due to the ovarian hyperstimulation. A preoperative diagnosis of heterotopic pregnancy was possible in only 26.3% of cases reported by Barrenetxea et al. [3]. In 73.7% of patients in this review, the correct diagnosis was achieved by surgical procedures. These results are not different from those reported by Tal et al. [14] referring to the 1971–1993 period. This means that improvements in ultrasound technologies have not been reflected in improvements in early diagnosis of heterotopic pregnancies. The difficulty of an accurate clinical and sonographic diagnosis in symptomatic patients clearly justifies a laparoscopy [3, 8, 13].

The main principle of the management of a heterotopic pregnancy consists of termination of the ectopic pregnancy without damaging the combined intrauterine pregnancy. The least invasive treatment should therefore be used to preserve the development of the intrauterine pregnancy [1, 3, 13].

Treatment of heterotopic pregnancies presents three problems: (1) serial human chorionic gonadotrophins (HCG) levels rise appropriately, (2) any treatment might compromise the coexistent intrauterine pregnancy and (3) the outcome of therapy cannot be followed by the traditional declining HCG levels. Serial sonography may follow size and shape of the ectopic pregnancy, but not trophoblastic activity [2].

Treatment of a cornual pregnancy can be either medical, surgical or expectant. In the medical management, potassium chloride can be used for the reduction of the ectopic gestation. One to 2 ml of 2 mEq potassium chloride can be injected into the ectopic gestational sac or foetal heart if the ectopic pregnancy is unruptured and clearly visualised. The contents can be aspirated after cessation of foetal cardiac activity. It is a minimally invasive procedure minimally jeopardizing the viable intrauterine pregnancy. Other drugs, like methotrexate with its potential adverse effects on the developing intrauterine fetus or prostaglandins with possible effects on uterine contractility, cannot be used for the reduction of the ectopic pregnancy in heterotopic pregnancies [3, 5].

During the last decades, surgical treatment of cornual pregnancies has shifted from the traditional laparotomy with cornual resection or hysterectomy to more conservative approaches including laparoscopic resection. The safety of laparoscopy during pregnancy has been well documented, and the advantages of laparoscopy over laparotomy in post-surgical recovery are well known [1–3]. Fertility results after laparotomy and after laparoscopy are similar [15].

Laparoscopy during pregnancy is best performed with the patient in the supine position. The lithotomy position may cause pressure on the legs, thereby increasing the risk of deep vein thrombosis. Extreme care must be taken when placing the insufflating needle and trocars to avoid injuring the uterus. An appropriate entry site for the Veress needle should be chosen, although in the first trimester the sub-umbilical entry can safely be used. The following trocars should be brought in under direct vision [1].

The different surgical techniques that can be used in the case of a cornual pregnancy are cornual resection or cornuostomy. Pre-tied ligatures (endoloop Ethicon®) or staplers can be used to achieve hemostasis of the proximal cornual region before resection. Others have described the achievement of hemostasis with ligation of the ascending branches of the uterine vessels [9].

In our case, we used the endoloop technique as described by Moon et al. [16] in the treatment of interstitial pregnancies. This technique provided us with an excellent tourniquet effect and resulted in effective hemostasis. Cornual closure was ended simultaneously with the removal of the conceptus by progressive tightening of the applied endoloop. This technique offered bleeding control during incision of the cornual area and both removal of the conceptus and effective closure of the uterine wall. This technique, however, can only be used in those cases where the cornual pregnancy is slightly elevated or pedunculated from the underlying myometrium. In those cases where the cornua is uniformly swollen and no distinction can be made between the cornual pregnancy site and the normal myometrium, a different technique should be used. A disadvantage of these conservative surgical techniques is that the cornual area might become weak, with the subsequent risk of uterine rupture during the ongoing pregnancy and labour. Therefore, in this case, we judged it safer to plan a primary caesarian section.

In the weeks after the corneal resection, we followed the patient very closely and were very vigilant for any signs of bleeding or rupture. Therefore, both we and the patient were easily alarmed, and the threshold for repeat laparoscopy was low. When the patient was admitted at 12 weeks and complained of the sudden onset of right fossa pain combined with vomiting, we decided to do a second laparoscopy. As this proved negative, we concluded that the abdominal pain was probably related to the vomiting.

The endoloop method is a simple, safe, effective and nearly bloodless technique for the treatment of selected cases of cornual heterotopic pregnancy. It offers a good prognosis for the ongoing intrauterine pregnancy. The possibility of uterine rupture should be discussed with the patient. Close follow-up is therefore crucial during the ongoing and subsequent pregnancies.

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