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## A viable caesarean scar pregnancy in a woman using a levonorgestrel-releasing intrauterine device: a case report

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### ABSTRACT

**Background:** Although the levonorgestrel-releasing intrauterine device (LNG-IUD) is one of the most reliable methods of contraception, it is associated with an increased risk of ectopic pregnancy in case of unintended pregnancy. A rare form of ectopic pregnancy is the caesarean scar pregnancy (CSP), with a high risk of serious maternal morbidity, such as uterine rupture, massive haemorrhage and resulting infertility. This report describes the first case of a viable CSP at 13 weeks of gestation in association with the use of a LNG-IUD.

**Case-presentation:** A 36-year-old Caucasian woman was referred to our outpatient clinic because of suspicion of a CSP. The pregnancy was unintended and was diagnosed during replacement of the LNG-IUD after five years. The patient had undergone two caesarean sections in the past. Ultrasound investigation showed an intact pregnancy of approximately 13 weeks of gestation located in the uterine scar. Because of the size of the gestational sac, a laparotomy was performed under general anaesthesia using a Joel-Cohen incision. The procedure was complicated by a total blood loss of 1500 mL, mostly caused by diffuse bleeding from the placental bed.

**Conclusion:** Unintended pregnancies in women using a LNG-IUD are frequently ectopic pregnancies with a preponderance to nidate outside the fallopian tube. Therefore, early diagnosis and location of the pregnancy in women using a LNG-IUD is essential.

### ARTICLE HISTORY

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### KEYWORDS

Levonorgestrel-releasing intrauterine device; LNG-IUD; caesarean scar pregnancy; ectopic pregnancy; Mirena

### Background

When properly situated, the levonorgestrel-releasing intrauterine device (LNG-IUD, Mirena<sup>®</sup>) is one of the most reliable forms of reversible contraception, with a pregnancy rate of less than 0.1% and a Pearl index of 0.06 [1,2]. The LNG-IUD is a plastic T-shaped device with the vertical stem containing a reservoir of 52 mg levonorgestrel and a polydimethylsiloxane elastomer mixture, covered by a polydimethylsiloxane membrane, which acts as a rate-limiting membrane releasing 20 mcg of levonorgestrel per 24 hours.

It is well-known that the use of an intra-uterine contraceptive increases the chance of an ectopic pregnancy [1]. However, in case of an unintended pregnancy, the chance of an ectopic pregnancy with an LNG-IUD is slightly higher than with copper IUDs [2]. Most of these pregnancies are located in the fallopian tube, but also in the ovary, the cornual part of the tube and the cervix. The most rare form of ectopic pregnancy is the caesarean scar pregnancy (CSP) [3,4]. Although rare, CSP can lead to serious complications such as uterine rupture or catastrophic haemorrhage, with a high risk of hysterectomy and even sometimes leading to maternal death [5,6].

We are the first to present a case of a vital CSP of 13 weeks of gestation in combination with a LNG-IUD.

### Case presentation

A 36-year-old Caucasian woman was referred to our outpatient clinic because of suspicion of a CSP. The pregnancy was unintended and was diagnosed during replacement of the LNG-IUD after five years. After replacement, the patient started bleeding excessively and an intact pregnancy was discovered on ultrasound. Beforehand, the patient had not experienced any abdominal symptoms or vaginal bleeding. The LNG-IUD was removed and the patient was referred. Her obstetrical history reported twice a caesarean section. Her further medical history was uneventful.

Ultrasound investigation showed an intact pregnancy of approximately 13 weeks of gestation (based on a biparietal diameter of 24 mm) located in the uterine scar. The placenta was located posteriorly.

After consultation with the patient, it was decided to evacuate the pregnancy. Because of the size of the gestational sac, a laparotomy was performed under general anaesthesia, using a Joel-Cohen incision. The gestational sac could already be visualized through the very thin, translucent uterine wall. An incision in the uterus was made cranial to this, and the fetus and placenta were removed. The margins of the uterine incision were excised to ensure complete removal of all pregnancy tissue and the uterus was closed in a single layer. Afterwards, tubal sterilization with



**Figure 1.** Transvaginal ultrasound six weeks post-surgery showed hardly any remaining dehiscence of the uterine scar.

Filshie clips was performed. The procedure was complicated by a total blood loss of 1500 mL, mostly caused by diffuse bleeding from the placental bed. The removed pregnancy tissue was sent for histological analysis, which showed a fetus without dysmorphic features and a normal placenta. After two days the patient was discharged from the hospital in good clinical condition.

Six weeks after the surgery, the patient revisited the outpatient clinic. Further recovery had been uneventful. Transvaginal ultrasound showed hardly any remaining dehiscence of the uterine scar (Figure 1).

## Discussion

The LNG-IUD has the lowest failure rate of the reversible contraceptive measures, with reported Pearl indices ranging from 0.1 to 0.6 and ectopic pregnancy rates ranging from 0.02 to 0.2 [1,2]. Causes of failure include unnoticed expulsion and dislocation of the device (not in the uterine cavity). Even when located in the cervix the device works and seems reliable [7], but in some cases pregnancy occurs despite adequate localization. In the latter cases, the relative risk of an ectopic pregnancy increases dramatically. In one prospective cohort study, 27% of all unintended pregnancies were ectopic [2]. Other studies report even higher ectopic pregnancy rates up to 50% [7–9]. Although most of these ectopic pregnancies are located in the fallopian tube, a larger percentage than usual seems located in the ovary, the cornual part of the tube or the cervix. So far, besides of tubal pregnancies, two cervical pregnancies, one cornual pregnancy and especially many ovarian pregnancies have been described in literature [10–12]. The preponderance of these aberrant locations in case of unintended pregnancy in combination with a LNG-IUD may be found in the working mechanism of this device. The contraceptive effect is most probably due to local endometrial suppression, thickening of the cervical mucus and alteration of uterotubal fluid, as well as suppression of ovarian function in some women. This makes it more likely that nidation may take place outside the endometrium and the fallopian tube.

The rarest form of ectopic pregnancy is the caesarean scar pregnancy (CSP), occurring in 1:1800 to 1:2226 of all pregnancies [3,4]. Little is known about the exact aetiology, however, it is thought that CSP occurs when a blastocyst implants on fibrous tissue within a wedge-shaped

myometrial defect at the site of a prior caesarean scar [13]. So far, only one CSP was described in combination with LNG-IUD use [14]. This pregnancy was terminated by medical treatment with methotrexate and leucovorin at five weeks of gestation, also because there was a heterotopic intrauterine pregnancy.

The first case of CSP was already reported in 1978, but over the past ten years there has been a significant rise in the number of reported cases. This could partially be due to the increasing number of caesarean sections, but also because of increased awareness and widespread use of first trimester ultrasound [3,4]. The risk increases with every additional caesarean delivery, with 50% to 72% occurring in women who had two or more caesarean deliveries. It is thought that with every added caesarean section more fibrosis occurs, enlarging the surface area of the scar and leading to poor vascularity, which impairs wound healing [13]. Most cases of CSP are diagnosed in first trimester by transvaginal ultrasound. Possible symptoms can be abdominal pain and vaginal blood loss, however, one third of patients is asymptomatic [13]. Early recognition is important because CSP can lead to severe complications such as uterine rupture and serious haemorrhage, needing hysterectomy with high maternal morbidity and loss of fertility [4,13]. Differential diagnosis includes cervical pregnancy and imminent spontaneous abortion [4–6]. Magnetic resonance imaging may be used when ultrasound is inconclusive [13]. Multiple treatment options for CSP have been described, such as surgical treatment, medical treatment, uterine artery embolization and curettage with or without simultaneous cervical cerclage. Because the condition is so rare, there are no universal guidelines and there is no consensus of the preferred mode of treatment. At least, first trimester termination of pregnancy is recommended to prevent serious complications and lower the risk of hysterectomy [5,13].

In our case, surgical evacuation through an open procedure was performed. This choice was made because of the size of the pregnancy and the risk of haemorrhage. Although serious haemorrhage occurred (total blood loss 1500 mL), there was no need to perform a hysterectomy.

## Conclusion

Unintended pregnancies in women using a LNG-IUD are frequently ectopic pregnancies with a preponderance to nidate outside the fallopian tube. Therefore, early diagnosis and location of the pregnancy in women using a LNG-IUD is essential.

## Consent for publication

Written informed consent was obtained from the patient for publication of this manuscript and any accompanying images.

## Disclosure statement

The authors declare that they have no competing interests.

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