CASE REPORT

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Discovery of a missing intrauterine system in the peritoneal cavity during cervical cancer surgery: a case report

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Abstract. Background: Intrauterine devices (IUD) are the most commonly used form of long-acting, reversible contraception; however, they can rarely be complicated by perforation at insertion or migration. Migration in the abdomen can cause inflammation, fibrosis, chronic pain and can affect neighboring organs but it can also be asymptomatic. Methods: We report the case of a woman with a story of a missing levonorgestrel-releasing intrauterine device who didn't undergo imaging to find the device and decided for the insertion of a second IUS. Then the missing device was discovered in her peritoneal cavity during surgery for cervical cancer. Results: The patient had at the same time the missing IUS in the peritoneal cavity and a correctly positioned IUS into the uterus and she had no symptoms related to the migrated IUS. Current literature reveals that up to 85% of patients with uterine perforation by IUD migration are asymptomatic. However, removal of the device is recommended, even in asymptomatic patients using minimally invasive methods if possible, including hysteroscopy, laparoscopy, cystoscopy, and colonoscopy depending on the location of the IUD. Conclusions: In a context of a missing IUD an abdominal imaging should be carried out to localize the device and its removal is recommended to avoid consequences. (www.actabiomedica.it)

Key words: intrauterine device, uterine perforation, migrated IUD, abdominal imaging

Introduction

Intrauterine devices/systems (IUDs/IUSs) are the most commonly used form of long-acting, reversible contraception (1), they are used by about 14% of women worldwide and up to 27% in some regions of the world (2). Copper- or levonorgestrel-based IUDs are among the most effective methods of contraception, with failure rates of less than 1% during the first 12 months of use (2). The mechanism of action of IUDs involves a foreign body reaction that leads to a sterile inflammatory response which is inhospitable to sperm and ova. Moreover, progestin-containing IUSs

have the additional benefit of causing a thickening of cervical mucus, making it more difficult for sperm to meet the egg and progestins can also cause decidualization of the endometrium and gland atrophy, both of which disrupt implantation (2). The benefits of IUD use include efficacy (>99% effective), lack of need for adherence, avoidance of exogenous estrogen, reversibility, and cost-effectiveness while the most common side effects are irregular bleeding and pain (2); however, available evidence suggests that pregnancy rates, adverse events, and discontinuation because of side effects during the first two years of IUD use are low and may not be clinically significant (3). Moreover, IUD use

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can rarely be complicated by pelvic inflammatory disease, contraceptive failure, expulsion, perforation at insertion, or migration (2). Perforation is exceptional but one of the most serious complications (1) and its rates reported in the literature range from 0.9 to 2.6/1000 insertions (4). Once the IUD migrates into the abdomen, it may cause inflammation and fibrosis, leading to adhesion formation and, less frequently, to chronic pain, intestinal obstruction or infertility. Organs such as the bladder or the bowel may be affected and perforation with acute or chronic sequelae has been reported (4). The clinical presentation of a patient with uterine perforation secondary to IUD placement can be either no symptoms or symptoms like abdominal pain and/or irregular vaginal bleeding (5); however, removal of a migrated IUD is recommended due to the risk of serious consequences if left in situ (4). Regarding the diagnostic methods, pelvic ultrasound is useful to confirm a migration of the IUD and abdominal X-ray has been used to confirm that the migrated IUD is in the pelvic/abdominal area rather than expulsed out through the vagina (5).

In this article, we present the first case of discovery of a levonorgestrel-releasing intrauterine system (LNG-IUS) in the peritoneal cavity during surgery for cervical cancer in an asymptomatic young woman with a story of missed IUS who didn't perform any radiologic imaging to verify the device's expulsion and who, in the meantime, underwent a second IUS insertion.

Clinical case

We present the discovery of a missing IUS Ky-leena in the peritoneal cavity during surgery for cervical cancer. The levonorgestrel-releasing intrauterine device (LNG-IUD) Kyleena, which indication is for the prevention of pregnancy, consists of a polyethylene T-shaped reservoir containing 19.5 mg levonorgestrel; over its effective 5-year lifespan it releases, on average, levonorgestrel 17.5 mcg/day (6). A 39-year-old, gravida 2, para 2 with 2 previous cesarean (in 2016 and 2018) was referred to our hospital because of a diagnosis of cervical cancer on cervical biopsy. She had no comorbidities and no abdominal surgery apart from 2 cesarean sections. The woman underwent IUS Kyleena

insertion for contraception on the 24th of May 2018, 46 days after the last cesarean section. At the check-up visit, after one month from insertion, the IUS strings were not found, and the 3D transvaginal ultrasound showed an image of dislocated IUS at the uterine isthmus. The gynaecologist tried but failed to remove the device in an outpatient setting and suggested a hysteroscopy. In October 2018 the patient underwent a diagnostic hysteroscopy that showed the absence of IUS in the uterine cavity. Because of the suspicion of spontaneous expulsion of the IUD the woman, together with her gynaecologist, in October 2019 decided the insertion of another IUS Kyleena and the following check-up visits were regular. In January 2023 the patient presented to our Center with a cervical biopsy diagnosis of HPV-related endocervical adenocarcinoma and was taken over by our gynaecological-oncology team. She underwent a transvaginal ultrasound that confirmed the correct position of the second-inserted IUS in her retroverted, 7 cm long, uterus and showed the presence of a 3 cm neoplasia involving the left posterior area of the cervix plus a left iliac internal lymphadenopathy (Stage T1b1). The isthmocele due to the previous cesareans was confirmed and adnexa were negative. In accordance with the cervical cancer guidelines (7), we submitted the patient to a deeper gynecological examination with cervical biopsies that confirmed the diagnosis and then the pre-operative TC-PET exam revealed, in addition to the intrauterine Kyleena (Figure 1), an imagine similar to a IUS in proximity to bowels (Figure 2). Due to the presence of the device in the abdomen the gynecological-oncology



Figure 1. PET-TC scan image of the correct positioned intrauterine IUS.



Figure 2. PET-TC scan image of the missing IUS in the peritoneal cavity.

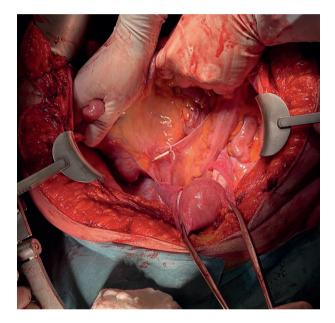


Figure 3. Intraoperative finding of the missing IUS located at the base of the mesentery at the caecum level.

team decided not to perform a staging MRI. For invasive-locally advanced HPV related cervical cancer stage T1b1 in patients with no pregnancy desire the guidelines suggest radical surgery performed by laparotomy (7). Therefore, in March 2023 she underwent radical hysterectomy, bilateral salpingo-oophorectomy, pelvic and lumbo-aortic lymphadenectomy with no residual tumor. During the mobilization of the small bowel the surgeon found the missing IUS encased at the base of the mesentery at the caecum level (Figure 3). The IUS was easily removed intact, and no organ perforations were found upon close inspection. The patient didn't complain of any symptoms, she was not

experiencing any abnormal or irregular bleeding (she had amenorrhea since the IUS insertion) and denied any pelvic or abdominal pain.

Discussion

IUDs are effective contraceptive measures, and although it carries an acceptable safety record, some complications may arise during its insertion or its long-term use; intra-abdominal IUD migration following uterine perforation is rare but in certain case serious complication (8).

Perforation usually occurs at the time of IUD insertion, but rarely can occur later. Risk factors for perforation include provider inexperience, retroverted uterus, immobile uterus, and myometrial defect from a previous cesarean delivery or myomectomy (1).

The mechanism by which IUD/IUS perforations occur is unclear and various etiologic theories exist. The first is that complete perforation occurs at the time the system is inserted, and the IUD/IUS is released beyond the serosa. The second is that the IUD/IUS is correctly placed into the uterus but is followed by transmural migration leading to perforation. The third is perforation on the site of a uterine scar of previous surgery (cesarean section, uterine myomectomy) (9).

Current literature reveals that many patients with uterine perforation by IUD migration may present with symptoms, but up to 85% are asymptomatic. In some cases, the diagnosis may arise by the appearance of clinical signs such as fever, abdominal pain, urinary tract infection or diarrhoea, or even serious complications such as occlusive syndrome or peritonitis due to bowel perforation. The clinical diagnosis is not always obvious, and further investigations are necessary to locate the device. Imaging has a great advantage in the topographic diagnosis of a migrated IUD. Abdominopelvic ultrasound is indicated as the first choice. It shows an empty uterine cavity or a para uterine IUD. Sometimes it does not find an IUD but cannot confirm a uterine perforation. Transvaginal ultrasound is a better way to assess uterine emptiness (2).

If the IUD is not seen by ultrasound, a simple abdominal X-ray can easily discern expulsion from migration. In specific complicated cases, in addition to

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transvaginal ultrasound and abdominal X-ray, a computed tomography (CT) scan or magnetic resonance imaging (MRI) scan of the abdomen and pelvis may provide more details about the exact location of the migrated IUD, assess for involvement of nearby organs and identify any associated visceral complication prior to surgical intervention (8). Once the dislocated device has been localized, the World Health Organization recommends surgical removal as soon as possible, even in asymptomatic patients. The recommendation is to use minimally invasive methods if possible, including hysteroscopy, laparoscopy, cystoscopy, and colonoscopy depending on the location of the IUD (2).

Although patients with a migrated IUD in the abdominal/pelvic cavity present often without symptoms, the rationale behind the removal of a misplaced IUD is mainly because of the risk of intra-abdominal adhesions formation, damage to adherent organs, chronic pelvic pain and infertility (5). For the migrated IUD in the peritoneal cavity, laparoscopic retrieval presents a minimally invasive approach and has been shown to be a safe and effective technique (5).

To enhance these statements, we introduce the following two studies involving a huge sample of patients.

The EURAS-IUD study of Barnett et al. that included 39,009 women with a newly inserted IUD (27,630 LNG-IUS users and 11,379 copper-IUD users) showed that approximately 25% of perforation cases were diagnosed more than 12 months after insertion. A small proportion of perforations were diagnosed during or immediately after insertion (2% of LNG-IUS and 17% of copper-IUD). Approximately 69% of perforations were diagnosed within 12 months of insertion, with the remaining perforations diagnosed between 1 and 5 years. In two LNG-IUS users, the perforation was diagnosed after the woman became pregnant. Most early perforations were detected following symptoms of pain and/or bleeding. However, in 29% of LNG-IUS and 17% of copper-IUD perforations, the perforation was asymptomatic. No perforation caused serious sequelae, such as bowel or bladder injury, septicaemia, or peritonitis and most IUDs that perforated the uterus were removed laparoscopically (66% of LNG-IUS and 59% of copper-IUD cases) (10).

An Analysis of Reports from Four National Pharmacovigilance Centres collected data from 701 cases of uterine perforation associated with the LNG-IUS Mirena that were reported to four national pharmacovigilance centres since the start of marketing to 15 July 2007. The results were the following: the mean time to detection was 306 days; in 47 of these cases (8.4%), the perforation was suspected or discovered at the time of insertion and in 143 cases (25.6%) the discovery took place after more than 1 year. In 102 cases (18%) uterine perforation was diagnosed during a control/check-up visit, for instance when the threads of the IUD were not visible. It was reported that the diagnosis of uterine perforation was confirmed by ultrasound in 302 of all reported cases (43%), by X-ray in 260 cases (37%), by CT in 47 cases (7%) and by MRI in 18 cases (3%) (11).

It should be highlighted that, in the specific clinical case here presented, the woman had at the same time the missing IUS in the peritoneal cavity and a correctly positioned IUS into the uterus. There are no articles available in literature revealing a story of two IUS inside a woman at the same time. Unfortunately, because of the disease of the patient, once the imaging revealed the presence of two devices in her body, we had no time to investigate her case (for example with blood test to evaluate her blood progesterone level) and she underwent surgery as soon as possible. For sure, it should be interesting to explore deeper this aspect if a similar case will occur.

Moreover, not only the IUD/IUS presents a risk of abdominal migration; in literature several cases of Essure Microinsert migration are described. Essure is a non-hormonal permanent contraceptive device which is trans-cervically implanted into the fallopian tubes and provides a sterilization alternative to hysterectomy or tubal ligation (12).

In the clinical case presented here the young woman was diagnosed with cervical cancer, however abdominal migration of intrauterine systems can occur also in other malignancies such as endometrial cancer and in many cases these young patients want to get pregnant; here the role of the gynecologist is very important to explain the patient the fertility preservation strategies in case of female cancer and to secure her the best fertility sparing treatment (13-16).

Conclusion

The IUD remains one of the safest and highly effective methods of contraception even if some complications occur during its use, one of which is uterine perforation with IUD migration in the abdomen. The diagnosis of a migrated IUD should be based on a thorough gynecologic analysis and appropriate radiologic imaging. Even if most of the patients remain asymptomatic, surgical removal is a first-line option to avoid serious complications; hysteroscopy or laparoscopy remains appropriate (2). The purpose of this paper is to emphasize that, in a context of missing threads or of IUS that is not identified in the uterine cavity by transvaginal ultrasound or hysteroscopy, abdominal imaging should be carried out to exclude vaginal expulsion and to localize the device in the abdomen in order to surgically remove it and avoid any related complication. Further investigation about displaced and migrated IUDs is needed, especially for women with a history of cesarean section or myomectomy.

Ethic Committee: Institutional review board of Udine. RIF. Prot IRB: 171/2023. Year: 2023.

Informed Consent Statement: Informed consent was obtained from all subjects involved in the study.

Conflict of Interest: Each author declares that he or she has no commercial associations (e.g. consultancies, stock ownership, equity interest, patent/licensing arrangement etc.) that might pose a conflict of interest in connection with the submitted article.

Authors Contribution: RS conceived the study, PA drafted the article, AM, DMM, SG, DL, VG made critical revisions to the manuscript. All authors listed have made a substantial, direct, and intellectual contribution to the work and approved it for publication.

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