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Case Report

A patient with a didelphys vaginal septum and infertility diagnosed by laparoscopy and magnetic resonance imaging

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Summary

Case Report: A 34-year-old woman presented with a desire to conceive. However, she could not participate in sexual intercourse because of severe vaginal pain. In vitro fertilization (IVF) was performed, and she achieved pregnancy. Although she could have had a twin pregnancy following IVF, one fetus was aborted. *Results:* Dilatation and curettage (D&C) was carried out with the patient under general anesthesia. The authors found a vaginal septum, which explained the severe vaginal pain upon attempting sexual intercourse. Although they carried out D&C, they were unable to completely remove the gestational sac. Diagnostic laparoscopy was then undertaken, revealing a duplicate uterus. *Conclusion:* This case indicates the need to be aware of the possibility of anomalies of the vagina and uterus when patients present with a disorder in sexual function. Clinicians should carefully listen to such patients' complaints in clinical practice.

Key words: Duplicate uterus; Infertility; In vitro fertilization; Laparoscopic diagnosis; Vaginal septum.

Introduction

The declining birth rate is currently one of the most serious social problems in the advanced world. However, an increase in the number of infertile couples is not widely recognized [1]. Therefore, assisted reproductive technology (ART) has been developed worldwide, and the numbers of infants conceived by ART have been markedly increasing. Congenital uterine anomalies arise because of abnormal development of the Müllerian duct during embryogenesis and have been associated with reduced fertility, miscarriage, preterm birth, and other adverse fetal outcomes [2-5]. The authors herein describe a case involving a patient with a duplicate uterus, vaginal septum, and infertility diagnosed by laparoscopy and MRI.

Case Report

A 34-year-old woman was referred to the outpatient clinic for her inability to become pregnant. She had a long history of an inability to participate in sexual intercourse because of severe vaginal pain. Otherwise, her medical history and familial history were not informative. Her husband's earlier semen examination had been normal. At this clinic, she underwent infertility screening including endocrinological tests. Physical examination with an S.S. Cusco vaginal speculum was abandoned because its insertion was blocked. Neither postcoital tests nor hysterosalpingography could be performed because they elicited severe pain and because of the opening limits of her hip joints. Although a bicornuate uterus was suspected based on a transvaginal ultrasonography (TV-USG) examination, a detailed examination was impossible because of her severe pain at each attempt.

Ovulation induction using clomiphene citrate and artificial insemination using her husband's semen [artificial insemination by husband (AIH)] was then performed because of her inability to have sexual intercourse and her desire for pregnancy. Prepared sperm suspensions were injected into the vagina, not into the uterus, to avoid pain. AIH in this form was carried out ten times unsuccessfully. She then underwent treatment by in vitro fertilization (IVF). Ovulation induction was performed using a long stimulation protocol, which involved administration of a gonadotropin-releasing hormone agonist and gonadotropin. Transvaginal follicular aspiration was performed with the patient under spinal anesthesia, and IVF was performed. Embryo transfer was conducted rapidly using an S.S. Cusco vaginal speculum because of her severe pain, but again she could not become pregnant. A second attempt at IVF with the long stimulation protocol was performed, and eight fertilized embryos were obtained. All were frozen. The third embryo transfer using two frozen–thawed embryos in a non-stimulated cycle was carried out, and she achieved pregnancy.

A 15.0-mm gestational sac (GS) was detected by TV-USG at five weeks of gestation. Another empty GS was detected by TV-USG at six weeks. At the same time, the fetus in the first GS was seen to have a crown–rump length of 4.0 mm and to have a fetal heartbeat (Figure 1a). However, at seven weeks, the patient experienced genital bleeding, and TV-USG showed disappearance of the fetal heartbeat. The other empty GS was also visualized in the uterus at that time (Figure 1b). The GS with the fetus had aborted completely, with only the empty GS remaining at eight weeks. The authors diagnosed a missed abortion. When the empty GS had not aborted at ten weeks of gestation, the authors planned dilatation and curettage (D&C) with the patient under general anesthesia. First, they attempted to insert a large Cusco vaginal speculum for the D&C, but it was blocked by a newly diagnosed vaginal septum (Figure 1c), which explained her pain and limited hip joint opening capacity. D&C was carried out with transabdominal ultrasonography guidance, but we were unable to completely remove the GS. Because the fundus of the uterus seemed to have an inflexible wall, it was difficult to smoothly perform the D&C. Diagnostic

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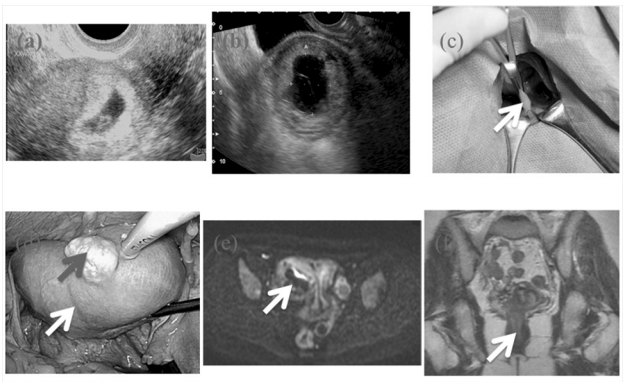

Figure 1. — (a) First gestational sac (GS) in the uterus, where a fetal heartbeat is detected. (b) The second GS is empty. (c) The white arrow indicates the vaginal septum. (d) Laparoscopic image: the white arrow indicates the boundary of two uteri and the red arrow indicates a uterine myoma. (e) MRI of the pelvic cavity showing two uteri. The white arrow indicates a hematoma in the uterus. (f) MRI of the pelvic cavity. The white arrow indicates the vaginal septum.

laparoscopy was then undertaken to avoid fundal perforation. The results of this surgery suggested the presence of a duplicate uterus (Figure 1d). MRI was carried out to evaluate the uterine malformation, which resulted in a diagnosis of a duplicate uterus with a vaginal septum (Figures 1e, f). Postoperatively, the patient was treated with methotrexate.

This study was approved by the Institutional Review Board of Asahikawa Medical University, Asahikawa, Japan. All procedures were followed in accordance with the ethical standards of the responsible committees on human experimentation (institutional and national) and with the Helsinki Declaration of 1964 and its later amendments. Informed consent was obtained from the patient to be included in the study. This article does not contain any study with animal participants that were performed by any of the authors.

Discussion

There were at least two major problems with this case. First, MRI should be performed to investigate the causes of anatomical or physical sexual disorders in their early stages. In recent studies, uterine anomalies were associated with vaginal anomalies in 45.2% of the patients evaluated [6, 7]. Therefore, the authors should have considered using MRI when the first TV-USG was performed. The second problem was that the operator for the two transvaginal follicle aspiration attempts, did not perform a sufficiently careful inspection. These two factors delayed the diagnosis.

The present authors normally perform vaginal embryo transfer following IVF to achieve pregnancy under circumstances in which the couple has had difficulty in conceiving naturally. In other cases, addressing and reversing sexual disorders allows the possibility of a natural pregnancy. A recent study showed that uterine anomalies do not affect pregnancy or live birth rates in women after assisted reproduction, but they do increase the preterm birth rate [8]. Several reports have indicated that one of the main causes of recurrent pregnancy loss is uterine malformation [9-11]. Clearly, further analysis of the possible link between uterine malformation and abortion rates in ART is needed.

In conclusion, this case has shown that the possibility of anomalies of the vagina and uterus must be addressed when patients present with a physical sexual disorder. Additionally, clinicians should carefully listen to such patients' complaints in clinical practice.

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