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Case Report

Fertility sparing treatment in an infertile patient with severe adenomyosis and a uterine fibroid: a case report

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ABSTRACT

Adenomyosis and uterine leiomyoma are benign lesions of the myometrium. Adenomyosis is a myometrial lesion characterised by the presence of ectopic endometrium with or without hyperplasia of the surrounding myometrium. Uterine fibroids, also called leiomyomas or myomas, are benign tumours that arise from the smooth muscle of the uterus. A 33-year-old nulliparous woman attended the clinic with primary infertility for 18 years. She was diagnosed with severe adenomyosis and uterine fibroids. She conceived successfully after undergoing *in vitro* fertilization (IVF) and a downregulated frozen embryo transfer (FET). Adenomyosis is a common cause of primary and secondary dysmenorrhea associated with infertility in young, nulliparous women. Fertility preservation is the primary goal in this population. Medical management is essential, and a properly downregulated and planned FET is the treatment of choice.

Keywords: Adenomyosis, Uterine fibroids, Leiomyomas, Infertility, Pregnancy, Case report

INTRODUCTION

Uterine leiomyoma and adenomyosis are benign lesions of the myometrium that are the major causes of irregular uterine bleeding that, in most cases, resolve after menopause.[1] Adenomyosis is a myometrial lesion characterised by the presence of ectopic endometrium with or without surrounding myometrial hyperplasia.[2] Uterine fibroids, also known as leiomyomas or myomas, are benign tumours that develop from the smooth muscle of the uterus. Although adenomyosis is a unique entity different from uterine fibroids, the developmental circumstances of these two diseases have a common basis that depends on the estrogen level.[3]

With a prevalence of 20-35% in women, adenomyosis manifests as abnormal uterine bleeding, pelvic pain, and abnormal enlargement of the uterus.[4] Age, multiparity, surgical disturbances of the endometrial-myometrial border, increased levels of both FSH and prolactin (PRL), smoking habits, and a history of depression are risk factors for adenomyosis.[2] Uterine fibroids can potentially cause infertility, recurrent pregnancy loss, and poor obstetric outcomes.[5] As a result, uterine fibroids and adenomyosis may require conventional treatment or surgery. The patient in this report was diagnosed with severe adenomyosis with uterine fibroids.

CASE REPORT

A 33-year-old woman and her 41-year-old husband visited our fertility clinic with complaints of primary infertility for 18 years, and they were both very anxious to conceive. She has complained of severe menorrhagia and dysmenorrhea for 10 years. Five years ago, three IUI cycles failed. Ultrasound revealed diffuse adenomyosis, a globally enlarged uterus (9-week size), and a 3×3 cm intramural fibroid in posterior wall. Her gynaecologist has suggested hysterectomy due to severe adenomyosis with 9-week-sized uterus. She visited our IVF clinic with above reports. On evaluation, her anti-mullerian hormone (AMH) level was 1.09 ng/ml. Her husband's semen analysis showed

severe oligoasthenoteratozoospermia (2 million/mL sperm count, 15% motility, and 2% morphology).

She has been counselled about IVF and intracytoplasmic sperm injection (ICSI). Proceeded with IVF. Ovaries were accessible using the trans abdominal approach. ICSI was performed on five eggs using the husband's sample. Three day 3 embryos were frozen.

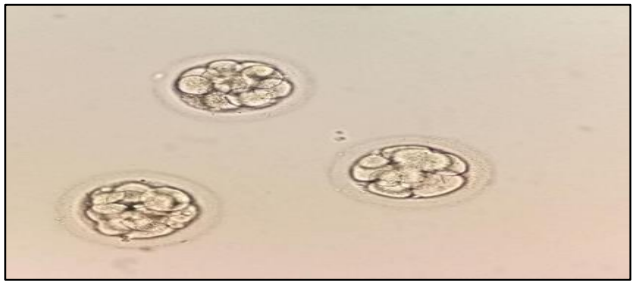

Figure 1: Day-3 frozen embryo.

She was downregulated with dinogest for 5 months followed She received three doses of gonadotropin releasing hormone (GnRh) *a*gonists (inj. luprolide 3.75 mg IM). FET is performed after downregulation. Her beta HCG was positive, and an ultrasound confirmed twin gestation. During her pregnancy, she was diagnosed with preeclampsia and treated with antihypertensives (tab. labetalol 100 mg 3 times a day).

Follow-up scans during pregnancy revealed that the first twin had unilateral Congenital talipes equinovarus (CTEV), also known as "club foot," and that the development of both twins was normal for their gestational age. She experienced P/V leaking at 35 weeks of pregnancy. Due to preterm premature rupture of membranes (PPROM), she underwent an emergency caesarian section. The first live twin preterm male baby was delivered cephalic at weight (2.02 kg), and the second live twin female baby was delivered breech at weight (1.850 kg). Intraoperatively, there was moderate postpartum haemorrhage (PPH) which was managed with uterotonics (Inj. carbetocin, oxytocin, and tranexamic acid). The postoperative period was uneventful.

DISCUSSION

Adenomyosis has previously been diagnosed through histologic analysis of hysterectomy specimens, leading to the assumption that it affects women in their 40s and 50s. However, with advancements in radiologic imaging, adenomyosis is being diagnosed at an earlier age.[6] The modern definition of adenomyosis was provided in 1972 by Bird, who stated, "Adenomyosis may be defined as the benign invasion of endometrium into the myometrium producing diffusely enlarged uterus, which microscopically exhibits ectopic non-neoplastic endometrial glands and stroma surrounded by hypertrophic and hyperplastic myometrium".[7]

Adenomyosis can be classified into two categories depending on the scope of lesion: 1. focal adenomyosis, in which the area of hypertrophic and distorted endometrium and myometrium is constrained (typically embedded within the myometrium), and 2. diffuse adenomyosis, which is the severe form of the disease and is distinguished by foci of endometrial mucosa (glands and stroma) scattered throughout the uterine musculature. The most common type is diffuse adenomyosis, while focal adenomyosis, especially on the cystic uterine glands, is rarer.[8] Here, the patient reported diffuse adenomyosis with a globally enlarged uterus (9-week size).

Recent research suggests an association between adenomyosis and infertility. While the mechanisms are mostly unclear, various hypotheses exist. By distorting the uterine cavity or destroying the uterine junctional zone, adenomyosis can disrupt healthy uterine peristalsis and uterotubal transfer. Endometrial function and receptivity would be compromised by an altered microenvironment, such as elevated local oestrogen levels and receptors with downregulated progesterone receptors. Thus, correcting the uterine environment by adenomyosis treatment may enhance pregnancy rates in infertile patients.[9]

Adenomyosis has been associated with poor pregnancy outcomes, an increased risk of preterm birth, preterm premature rupture of membranes (PPROM), and foetal growth restriction (FGR).[10] Furthermore, adenomyosis has been considered a cause of repeated implantation failure. Adenomyosis is thought to cause inflammatory changes in the endometrium and adversely affect the composition of endometrial fluid**,** both of which can impair implantation.[11]

IVF treatment can be combined with medical therapy to increase pregnancy rates. Due to the detrimental effects of adenomyosis on IVF outcomes and the iatrogenic complications associated with controlled ovarian stimulation (COS), segmented-IVF with FET should be considered.[9] To control symptoms associated with adenomyosis, a number of hormonal therapies are currently being used, such as progestin, gonadotropin-releasing hormone analogues (GnRHa), and oral contraceptives.[12] In this case, the patient was downregulated with gonadotropin-releasing hormone analogues (GnRHa) and dienogest (oral progestin)

Adenomyosis and leiomyomas usually coexist.[2] In our case, the patient presented with diffuse adenomyosis with a uterine fibroid.

Uterine fibroids are the most common type of benign uterine tumor. They are myometrial monoclonal tumours of the smooth muscle of the uterus. The cause of leiomyomas in adolescents and adults is unknown; however, they are known to develop in response to both oestrogen and progesterone stimulation, and their prevalence rises during the reproductive years and drastically reduced after menopause. Obesity, pregnancy,

early menarche, and exposure to exogenous oestrogen are common factors that affect fibroid development .[13]

Because the majority of women with myomas are asymptomatic, the incidence of undiagnosed uterine fibroids is high. Women who are symptomatic are more likely to experience pelvic pain, infertility, pregnancy complications, and excessive uterine bleeding, which can lead to anaemia. Early and late miscarriage, preterm birth, foetal malpresentation, placental abruption, post-partum haemorrhage, and an increased risk of caesarean delivery are all obstetric complications associated with fibroids. Myomas might induce compression-related symptoms such as dyspnoea, frequent urination, or gastrointestinal problems when they expand. Extremely large myomas can cause major consequences such as respiratory failure due to diaphragmatic compression or an incarcerated abdominal wall hernia.[14,15]

Uterine leiomyoma and adenomyosis are considered estrogen-dependent; these benign diseases have similar symptomatology and reproductive consequences.[1] Adenomyosis is often misinterpreted as uterine fibroids because the symptoms are so similar. In terms of ultrasound scan differentiation between leiomyoma and adenomyosis, the former is associated with defined margins, round lesions causing mass effects with calcification, and peripheral vascularization. Adenomyosis, on the other hand, is distinguished by lesions of varying shapes with poorly defined margins and no calcification. Along with the myometrial hyperplasia, there is a rectilinear pattern of vascularization.[11]

CONCLUSION

In conclusion, this case highlights an uncommon but little-known factor that can affect young nulliparous women and cause both primary and secondary dysmenorrhea. Preserving fertility is the main goal of this population. Avoid hysterectomy as a treatment approach for infertile women. Medical management is crucial in the management of an infertile adenomyosis patient. A proper, downregulated, and planned FET is the treatment of choice for severe adenomyosis patients.

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