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Case Report

Subfertile Woman with Painful Umbilical Nodule: A Case Report

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Abstract

Primary umbilical endometriosis is very rare.Painful umbilical nodule in a sub fertile woman can clinically be diagnosed as umbilical endometriosis. The treatment is surgical excision of umbilicus but subfertility is difficult to manage.

Keywords: Umbilical endometriosis, Infertility, Endometriosis.

INTRODUCTION

The umbilical nodule can occur due to various diseases. About 38% of umbilical nodules are benign lesions of the umbilicus, 32% due to endometriosis and 30% due to metastatic disease from a distant location [1]. A variant of cutaneous endometriosis, umbilical endometriosis was first described by Villar in 1886 [2]. So occasionally it is called Villars nodule. Umbilical endometriosis comprises 6.5-1% of all cases of extra genital endometriosis [3].

Umbilical endometriosis secondary to surgical, commonly laparoscopic, scar is usual. Primary umbilical endometriosis is very rare. The theories that describe its mechanism include intraperitoneal migration of endometrial cells during menstruation, coelomic metaplasia or metastatic dissemination of endometrial cells by blood vessels and lymphatics [4].

CASE PRESENTATION

A 30 yearold nulliparous woman presented with a history of cyclical umbilical pain, and a tender nodule in the umbilicus, enlarging in size for the past 6 months. The patient hasa history of progressive dysmenorrhea characteristic of endometriosis for 1 year and a history of infertility for 2 years.

According to the patient'sstatement, she has cyclical menstruation with average flow and duration since her menarche. Initially her menstruation was painless but she developed progressive secondary dysmenorrhea for the last 1 year. She has been trying to conceive for the last two years since her marriage. With the complaints of infertility and dysmenorrhea, the woman consulted with the outpatient department of Reproductive Endocrinology and Infertility of BSMMU several times andwas diagnosed as a case of right ovarian endometrioma. She got ovulation induction with letrozol followed by sonographic monitoring and HCG trigger for 2 cycles. She noticed cyclical pain and swelling in her umbilicus for the last 6 months. Shewasadvised hospital admission for surgery.

She had no past history of surgery and her medical history was unremarkable.Her husband is a 26 year old service holder by occupation, smoker, nondiabetic, normotensive, havinga recent normal semen analysis report. She had no history of TB, mumps, or any pelvic surgery.

On physical examination, she had a soft, tender swelling in the umbilicus, bluish in color measuring approximately 1 cmx1 cm, extending into the deeper tissues.Unlike umbilical hernia, it is

irreducible by gentle digital pressure and cough reflexis negative (Figure-1). There was no sign of infection and bleeding in the umbilical lesion. The patient had a calculated body mass index of 24 kg/m^2. Vaginal examination revealedthat cervixwas apparently healthy, both forniceswere full and tender. A mass was adherent to the uterus withrestricted mobility. On digital rectal examination, rectal mucosa was free, no deposit in cul de sac. Transvaginal ultrasound findings were bilateral chocolate cysts measuring about (5.5×5.1) cm in right ovary and (2.3×2.1) cm in left ovary.

Based on history, clinical examination and investigation the provisional diagnosis was made. The provisional diagnosis was primary subfertility with bilateral endometrioma with umbilical endometriosis.

After evaluating history, physical examinations and lab investigations patient underwent diagnostic laparoscopy which revealed endometriomas in both ovaries, obliteration of cul de sac in right side and adhesion with intestine. Cystectomy and adhesiolysis were performed, trying to restore tubo-ovarian relationship. Both fallopian tubes were patent on instillation of methylene blue. Surgical excision of the umbilicus and its surrounding tissues consisting of skin, fat, and fascia and reconstruction was performed (Figure 2-4). Reconstruction was done by using a purse-string suture followed by fixation to rectus sheath (Figure 5).

Fig-1: Bluish umbilical mass or nodule

Fig-2: Periumbilical incision with free margin

Fig-3: Wide excision of umbilical endometriosis

Fig-4: Omphalectomy

Fig-5: Reconstruction of umbilicus

Surgery was the effective treatment for the woman. As patient had associated umbilical endometriosis & endometrioma of size more than 5 cm, severe dysmenorrhea with prolonged subfertility, so surgical treatment was considered to be more effective for her. We could assess tubal patency in the same sitting. Anatomical restoration of tubo ovarian relationship could be done only by surgery.

Histopathological examination of the specimens revealed umbilical tissue with endometriosis consisting of endometrial glands and stroma, and cyst wall compatible with endometrioma consisting of endometrial glands and stroma.

The definitive treatment of primary umbilical endometriosis is surgical excision with free margin. As the patient had bilateral endometrioma, the size of the endometrioma was more than 5cm, medical treatment was likely to delay fertility outcome and the patient's age was 30 years, the definitive treatment was laparoscopic cystectomy with chromopertubation and excision of umbilical endometriosis.

As the patient was 30 years of age, had advanced endometriosis with chocolate cysts, umbilical endometriosis, and history of primary subfertility, conservative management, compared to surgery would not be effective. Conservative treatment with hormones helps improve symptoms of dysmenorrhea but concomitant ovarian suppression adversely affects fertility.

The next plan of managementfor the woman was down-staging with GnRH for 3 month and IVF (in vitro fertilization). But the patient was economically poor. She was not capable for IVF. So, we expected to do ovarian stimulation with gonadotropins followed by intra-uterine insemination for 3 cycles. But before that we would do serum AMH to assess ovarian reserve.

DISCUSSION

Benign lesions of umbilicus include umbilical hernia, lipoma, dermoid cysts, dermal nevi, congenital malformation of urachus, foreign body granuloma etc [2, 5]. Umbilical hernia appears typically as a lump around the umbilicus. Pain is the most common complaint for which the surgery of repair is done. The hernias appear above or below the umbilicus through a weak area in linea alba. The herniation is due to increasing intra-abdominal pressure. The hernia sac may contain pre-peritoneal fat tissue, omentum and intestine.[6] Sister Mary Joseph nodule is the sequelae of malignant metastasis from tumor in the pelvis or abdomen. The nodule typically appears in umbilical or para-umbilical areas,varying in size between 0.5-15 mm, has a firm consistency, occasionally painful and discharging fluid.It is associated with primary neoplasm of gastrointestinal origin (35-65%), genitourinary tract (12-35%), lung cancer, lymphoma and other intra-abdominal and pelvic malignancies. The source cannot be identified in upto 30% of patients [7].

The diagnosis of umbilical endometriosis is primarily clinical. The patients are in reproductive age group and presents commonly with swelling, pain, discharge or cyclical bleeding from the umbilicus. Bluish black in color, the lesion enlarges, and becomesmore painful, and bleeds about the time of

menstruation [8]. However symptoms may not be associated with menstrual cycle in about 25% of the patients [2].

Multiple radiologic diagnostic procedures like ultrasound, CT scan or MRI have been used for diagnosing umbilical nodules. But none of the radiologic studies is specific to the diagnosis of umbilical endometriosis [2].

Preoperative medical therapy with continuous contraceptive pills, progesterone or GnRH agonists may be used to provide symptomatic relief and to reduce the size, but surgical removal with clear margin is the appropriate treatment. Simultaneouslaparoscopy to diagnose and treat pelvic endometriosis is indicated in patients with symptoms and infertility [2].

Omphalectomy with concomitant laparoscopic approach allows radical excision of the umbilical endometriosis. Following the laparoscopic procedure the trocar is moved into the right axillary areas. Under direct vision the previous peri-umbilical incision is completedtrans-cutaneous to include the fascia and peritoneum [5].

According to the European Society of Human Reproduction and Embryology (ESHRE) guideline for endometriosis, women with endometrioma who suffer from infertility and pelvic pain, should have surgery for excision of endometriotic cyst. The other surgery that can be done is simple drainage followed by electrocoagulation of cyst bed. Postoperative spontaneous pregnancy rate is higher following cystectomy. Both procedures carry the risk of losing ovarian reserve, because of unintended removal of ovarian cortical tissue or thermal damage [10-12].

The gold standard of diagnosis of endometriosis is laparoscopy. Greater magnification and better illumination allow identification of endometriotic lesions and their extent of involvement. Visual diagnosis followed by surgical excision or ablation and adhesiolysis of endometriotic lesions is effective in reducing pain.

Chronic pelvic pain related to severe endometriosis can be managed by laparotomy as well as laparoscopy [10]. Laparoscopic surgery, in terms of excision or ablation of endometriotic lesions is better than diagnostic laparoscopy alone in providing the outcomes of pain relief, clinical pregnancy rate and live birth [11].

Ovarian reserve is likely to decrease following laparoscopic cystectomy. If it is below 1 ng/dl success rate is very poor.[12] According to ESHRE guideline 2018, GnRH agonists can be prescribed for a period of 3 to 6 months prior to in vitro fertilization to improve clinical pregnancy rates in infertile women with endometriosis.

The success rate of IVFis relatively poor in endometriosis.A meta-analysis of 22 published studies reveals that following in vitro fertilization, the chance of pregnancy is significantly lower (odds ratio 0.56, 95% confidence interval 0.44-0.9) in women with endometriosis when compared to controls with tubal factor infertility. In women with endometriosis, there are significant decrease in the number of oocytes retrieved as well as in the rate of fertilization and implantation [13-19].

CONCLUSION

Painful umbilical nodule in a sub fertile woman can clinically be diagnosed as umbilical endometriosis. The treatment is surgical excision of umbilicus but subfertility is difficult to manage.

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