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Massive uterine adenomyosis: a long-term followup of its conservative surgical treatment

Alessandro Bulfoni³, Giada Frontino⁴, Daniela Alberico¹, Stefano Bianchi², Luigi Fedele¹

¹ Department of Obstetrics and Gynecology, Fondazione IRCCS Ca' Granda Ospedale Maggiore Policlinico, Mangiagalli via Francesco Sforza 35, Milan 20122 Italy

² University of Milan, Ospedale San Giuseppe, via San Vittore 12, Milano

³ University of Milan, Humanitas Pio X, via A. Manzoni 56, Milano

⁴ Consultant Obstetrician and Gynecologist London

ABSTRACT

Fifteen patients with massive diffuse adenomyosis underwent laparotomic uterine debulking. Retrospective analysis regarded menstrual characteristics, pelvic pain, dyspareunia and reproductive outcome. Statistical analysis was obtained using t test. $P < 0.05$ was considered significant. Mean age was 34.6 years.

Follow-up lasted a mean of 41.4 months. No intraoperative nor postoperative complications occurred. At 12 months after surgery, 13 patients (87%) reported complete resolution of menorrhagia, mean VAS for dysmenorrhea was 1.8 ± 2.48 and mean VAS for chronic pelvic pain was 1.0 ± 2.17 . At 6 months after surgery all patients had normal hemoglobin levels. One patient had 2 spontaneously-conceived term pregnancies. Despite the low postoperative pregnancy rate obtained in this study, symptoms were significantly improved in terms of dysmenorrhea, menorrhagia and sideropenic anemia. The long-term follow-up shows persistence of the improvement in symptoms.

Keywords: diffuse adenomyosis, conservative surgery, dysmenorrhea, menorrhagia.

SOMMARIO

Quindici pazienti con adenomiosi diffusa sono stati sottoposti a debulking uterino laparotomico. L'analisi retrospettiva ha osservato le caratteristiche mestruali, il dolore pelvico, la dispareunia e l'esito riproduttivo. L'analisi statistica è stata condotta utilizzando un t test. $P < 0.05$ è stato considerato come significativo. L'età media dello studio è di 34.6 anni. Il follow-up è durato in media 41.4 mesi. Non si è verificata nessuna complicanza, né durante l'operazione né dopo.

A 13 mesi dall'operazione, 13 pazienti (87%) hanno riportato una completa risoluzione della menorrhagia. La media della VAS per la dismenorrea è stata di 1.8 ± 2.48 mentre la media della VAS per il dolore pelvico cronico è stata di 1.0 ± 2.17 . A 6 mesi dall'operazione tutti i pazienti hanno riportato livelli di emoglobina regolari. Una paziente ha avuto 2 gravidanze spontanee a termine. Nonostante il basso tasso di gravidanza post-operatorio ottenuto in questo studio, i sintomi quali: la dismenorrea, la menorrhagia, e l'anemia sideropenica, sono migliorati significativamente. Il follow-up a lungo termine mostra un continuo miglioramento dei sintomi.

Corresponding Author: Luigi Fedele luigi.fedele@unimi.it

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INTRODUCTION

Uterine adenomyosis is a common benign gynecologic disorder which is commonly observed in women in their late 30s and 40s. Symptoms are nonspecific, can frequently be severe and include menorrhagia, dysmenorrhea, chronic pelvic pain and infertility⁽¹⁾. While the focal subtype of adenomyosis is usually treated conservatively through resection of the nodules, hysterectomy is still considered the standard treatment for diffuse adenomyosis. In contrast with endometriosis^(8,9), uterine adenomyosis is more frequently found in multiparous women, but it also affects nulligravid women. Treatment in the latter cases is therefore guided principally by the patient's desire to preserve the uterus and potential fertility and to avoid morbidity linked to hysterectomy. Management of diffuse disease is still lacking specific guidelines, due to the paucity of studies in the literature especially regarding its conservative treatment. The present study reports the long-term follow up of 15 patients who have undergone conservative surgical treatment for massive diffuse uterine adenomyosis.

MATERIALS AND METHODS

All fifteen patients with intraoperative and histological confirmation of massive diffuse adenomyosis involving at least two-thirds of the anterior and/or posterior uterine walls, who underwent laparotomic conservative uterine debulking by the same surgeon (L.F.) between November 2004 and April 2010 at the tertiary center of the Department of Obstetrics and Gynecology of our Institute, were retrospectively analysed regarding menstrual characteristics, pelvic pain, dyspareunia and reproductive outcome. Massive diffuse adenomyosis was defined at the preoperative workup as the presence of lesions involving at least two-thirds of the uterine body. Preoperative work-up included a complete blood panel, transvaginal and transabdominal pelvic ultrasound (US) and pelvic magnetic resonance imaging (MRI) (**fig.1**). Laparotomic uterine debulking involved resection of the area of myometrium affected by adenomyosis, judged by macroscopical visual inspection and palpation following myometrial incision of the diseased area suggested at the preoperative imaging workup.

Gonadotropin-releasing hormone agonists were not administered preoperatively in order to allow adequate intraoperative evaluation of the consistency and size of the adenomyotic mass.

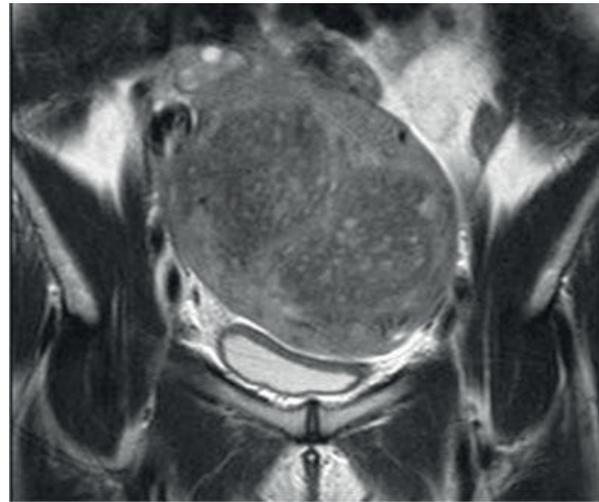


Figure 1.
T2-weighted fat-saturated MRI image showing transverse section of uterine myometrium subverted by diffuse adenomyosis.

Oral iron supplementation was prescribed with 329,7 mg ferrous sulphate and 4 micrograms folic acid for 4 months prior surgery and 1 month postsurgically. Surgery was performed through a suprapubic transverse laparotomic incision, followed by uterine mobilization and pelvic exploration. The uterus is kept lifted out of the abdomen during the surgical procedure. After inspection and palpation of the uterus body, a sagittal incision is made on the anterior and/or posterior walls so that the affected myometrium is exposed. The endometrial cavity is opened systematically in order to aid excision of the adenomyotic lesions while sparing the endometrium. Once the extent of the diseased myometrium has been assessed, a monopolar knife is used to excise the margins of the adenomyotic tissue (**fig.2**) while taking care to avoid damage to the Fallopian tubes.



Figure 2.
Cuneiform section of myometrium affected by diffuse adenomyosis.

Resection was considered to be complete when at least two-third of the adenomyotic tissue had been removed while leaving enough myometrium to allow adequate myometrial reconstruction. The endometrial cavity is closed with interrupted sutures in 3-0 Vicryl. The myometrial defect is then sutured along two layers in 2-0 Vicryl, respectively a first deep interrupted sutures then followed by a superficial continuous suture which includes the serosa.

Follow up visits were programmed at 6, 12, 24 months and every year thereafter. Parameters that were evaluated at each follow up visit included a blood panel, a pelvic US scan and symptom analysis, i.e. dysmenorrhea, dyspareunia, pelvic pain and menstrual bleeding. Dysmenorrhea, dyspareunia and pelvic pain were analyzed using a Visual Analogue Scale (VAS)⁽²⁾. Statistical analysis was obtained using a t- test. $P < 0.05$ was considered significant. Menstrual bleeding was referred by the patients as mild, moderate or severe. Reproductive outcome was analyzed in those patients seeking a pregnancy.

The study was exempt from Institutional Review Board approval due to its retrospective nature of analysis of a conservative surgical treatment.

All women included in this study had signed an informed consent agreeing for their clinical data to be used anonymously for research purposes, along with the informed consent regarding the surgical procedure.

RESULTS

Mean patient age was 34.6 (26-41) years. Follow up lasted at least 19 months (mean 41.4 months; 19-38 months). All patients preoperatively reported moderate-severe dysmenorrhea with a mean VAS of 7.5 ± 1.46 ($P < 0.0001$). Six patients (40%) complained of deep dyspareunia, two of which mild and four moderate, with mean VAS of 2.1 ± 2.74 ($P < 0.04$). Eight of the 15 patients (47%) reported chronic pelvic pain, two of which mild and six of which severe, with mean VAS of 3.6 ± 3.85 ($P < 0.031$). The other seven patients complained of chronic pelvic pressure.

Four and 4 patients had respectively primary and secondary infertility. One patient had previously undergone surgery for diffuse adenomyosis. Menorrhagia was complained by all 15 patients. The mean preoperative hemoglobin levels showed sideropenic anemia in 13 of the 15 patients (87%), despite all had been taking oral

iron supplements for 4 months prior to surgery; 7 and 6 of these had respectively moderate and mild anemia. In 5 patients the area of adenomyosis involved the posterior uterine wall, in 4 both the anterior and posterior walls, in another 4 the anterofundal portion of the uterus, and in 2 only the anterior wall. Surgery lasted a mean time of 108 minutes (89-145 minutes) and blood loss was a mean of 660 mL (50-2000 mL). None of the patients required blood transfusions and no major intra- nor postoperative complications occurred. Histopathological examination confirmed uterine adenomyosis in all 15 patients. (Table 1).

Table 1.

Table 1. Baseline characteristics of the cohort.

Characteristics	Number
Age (years)	34 (26-41)
Dysmenorrhea moderate-severe	15 \pm 0
Deep Dyspareunia	6 (40%)
- Mild	2
- Severe	4
Chronic pelvic pain	8 (47%)
- Mild	2
- Severe	6
Infertility	
- Primary	4
- Secondary	4
Menorrhagia	15 \pm 0
Preoperative sideropenic anemia	13 (87%)
- Mild	6
- Moderate	7
Site of uterine adenomyosis	
- Posterior wall	5
- Anterior e posterior wall	4
- Anterofundal wall	4
- Anterior wall	2

At 12 months after surgery, 13 patients (87%) reported complete resolution of menorrhagia, while 2 referred slight improvement, i.e. moderate menorrhagia. Postoperative data at 12-month follow up showed absence of dysmenorrhea in 9 cases (60%), mild and moderate dysmenorrhea in respectively 3 cases and moderate in another 3 (mean VAS 1.8 ± 2.48).

Of the 6 patients who preoperatively referred dyspareunia, 4 (67%) referred its complete remission and only 2 (33%) referred mild dyspareunia (mean VAS 0.4 ± 1.06). Five of the eight patients (62.5%) who preoperatively complained of chronic pelvic pain referred

absence of pain since after surgery, 2 had moderate pain and 1 mild pain (mean VAS 1.0 ± 2.17). At respectively 6 and 12 months after surgery, all patients had normal hemoglobin levels. Mean age of the patients wishing to conceive was 37 years (30-44 years). Only one patient, who had primary infertility, had 2 spontaneously- conceived term pregnancies, which resulted in programmed caesarean sections. One other patient with primary infertility unsuccessfully attempted three intrauterine inseminations. One patient with secondary infertility also unsuccessfully underwent one cycle of in-vitro fertilization and embryo transfer (IVF- ET). Respectively two patients with primary infertility and two with secondary infertility are currently being evaluated at our Infertility Center. Four patients refer they will seek a pregnancy in the next future. (Table 2).

Table 2.

At 12 months after surgery.

Characteristics	Number
Absence of Dysmenorrhea	9 (60%)
Mild Dysmenorrhea	3
Moderate Dysmenorrhea	3
Absence of Dyspareunia	4 (67%) *
Mild Dyspareunia	2 (33%) *
Absence of Chronic pelvic pain	5 (62.5%)*
- Mild	1
- Moderate	2
Menorrhagia	13 (87%)
Postoperative sideropenic anemia	0
Mean age of the patients wishing to conceive	37 (30-44)

* four of the six that referred dyspareunia preoperatively

* five of the eight that referred chronic pelvic pain preoperatively

DISCUSSION

Surgical therapy in women with severe diffuse adenomyosis has been limited to hysterectomy for many years. With the continuous development in imaging methods, uterine adenomyosis not only is encountered more frequently, but may also be more accurately described in the growing number of nulliparous patients over 35 years of age seeking a pregnancy. Despite such growing diagnostic power, there are scanty reports in the scientific literature on the therapeutic options in women with diffuse symptomatic adenomyosis. Data is even scantier on the management of most severe, yet not uncommon, forms of

massive diffuse involvement of the myometrium. Symptoms in these cases are often severe as well as refractory to the commonly recommended hormonal treatments. In infertile and/or symptomatic patients seeking a pregnancy, a diagnosis of diffuse adenomyosis may only temporarily benefit from medical therapy such as the levonorgestrel intrauterine device (Lng- IUD) or gonadotropin releasing hormones agonists, whereas the recommended surgical treatment, i.e. hysterectomy, cannot be considered a treatment option in these patients.

Among the conservative surgical treatments for uterine adenomyosis, new and less aggressive options have been introduced including endomyometrial ablation and uterine artery embolization. However, the scientific literature reports suboptimal efficacy of these methods, i.e. around 50% in the treatment of diffuse disease^(3,4). Also, most studies evaluating these methods lack histopathologic confirmation of diffuse adenomyosis and have a relatively short follow up, thus not allowing full assessment of the long-term benefits.

Above all, the number of cases of diffuse uterine adenomyosis are scanty and its myometrial extension superficially described nor stratified. While these minimally invasive techniques may be effective in the focal type of adenomyosis, accurate demarcation nor complete removal of diffuse lesions cannot be achieved. On the other hands laparotomy also enables palpation and identification of diseased tissue, easy hemostasis and assessment of the integrity of the endometrial cavity.

Similar to the only two reports in the literature that studied conservative treatment in cases with massive diffuse uterine adenomyosis, this study is flawed by its nonrandomized nor controlled design. In spite of these shortcomings, its methodology does show patient stratification, a long-term follow up and the presence of histopatological diagnosis, as well as uniformity of the type of adenomyosis. While in the by Osada and colleagues⁽⁵⁾ 14 out of 26 (53,8%) patients obtained a term pregnancy, in our study post treatment pregnancy rates were low alike the study by Nishida et al.⁽⁶⁾ (1 out of 44 cases). In our study, 4 (67%) of the six pregnancy-seeking patients were aged between 36-44 years, which may have influenced the obstetric outcome. Unlike our study, Nishida et al.⁽⁶⁾ performed monolateral salpingectomy systematically in all patients as part of the surgical procedures.

The number of patients included in this

study is admittedly low, due to the stringent inclusion intraoperative criteria in which only cases of diffuse adenomyosis involving at least two-thirds of the anterior and/or posterior walls were included, thus yielding more specific data. Also other studies in literature presents a small number of patients. Despite the low postoperative pregnancy rate obtained in this study, symptoms were significantly improved in terms of dysmenorrhea, menorrhagia and sideropenic anemia. The long-term follow up also shows persistence of the improvement in symptoms, thus providing continuous relief in terms of quality of life of this patients.

Various medical and surgical methods have been developed for the management of a

condition that until recently had hysterectomy as its only option. Data in the literature especially concerning conservative treatment for extensive diffuse uterine adenomyosis is extremely scanty, thus further research on these treatment methods is warranted⁽⁷⁾. This study shows that conservative treatment of massive diffuse adenomyosis can be considered a safe as well as effective procedure, with good results in terms of relief from severe dysmenorrhea, pelvic pain and menorrhagia.

DECLARATION OF CONFLICTING INTERESTS

The author(s) declared no conflict of interests.

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