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Review Article

Uterine-sparing surgery for adenomyosis and/or adenomyoma☆

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ABSTRACT

Adenomyosis of the uterus is defined as the presence of endometrial tissue, including glands and stroma, situated at least 2.5 mm below the endometrial–myometrial junction and widely distributed within the myometrium layer of the uterus. There is no consensus on the appropriate treatment for symptomatic uterine adenomyosis in women who want to preserve their uterus, partly because adenomyosis is somewhat enigmatic in diagnosis and owing to its clinical significance. Hysterectomy, through either exploratory laparotomy or minimally invasive procedures, is a definite treatment for uterine adenomyosis, once the women have completed childbirth or do not require future fertility. However, many women with a uterine pathology still have a strong desire to preserve the uterus, for which conservative and uterine-sparing procedures are increasingly used, and with which fertility preservation or quality-of-life improvement can be achieved. Although medical management can be effective, similar to the management of uterine fibroids (myoma), its effect is often transient and rapid regrowth of adenomyosis and relapse of symptoms and signs always occur once the treatment is stopped. Therefore, other strategies should be selected. Conservative and uterine-sparing surgery might be one of the most familiar procedures of these uterine-sparing procedures. In this article, the latest knowledge and research evidence on uterine-sparing surgery for uterine adenomyosis are reviewed.

Introduction

Adenomyosis was first described in 1860 by von Rokitansky, who found a hysterotropic occurrence of islands of endometrium scattered throughout the myometrium [1]. Frankl subsequently specified it as adenomyosis interna in 1925 [2]. During the last decade, adenomyosis was identified as a downgrowth and invagination of the endometrial basalis into the adjacent myometrium after disruption of the normally intact boundary between them [3]. Adenomyosis is characterized by the presence of heterotopic endometrial glands and stroma within the myometrium, accompanied with a variable degree

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of adjacent myometrial hyperplasia [4]. By definition, adenomyosis causes globular and cystic enlargement of the myometrium, with some cysts filled with extravasated, hemolyzed red blood cells and siderophages in gross appearance. In adenomyosis, the ectopic endometrium is located past the "last" glands of the basalis, and circumferentially surrounded by bundles of hypertrophic smooth muscle cells (collar), which clearly differ cytologically from stromal fibroblasts, and foci of adenomyosis (proliferative types of adenomyotic glands and stroma) seen more than 2.5 mm deep in the myometrium or more than one microscopic field at 10× magnification from the endometrium–myometrium junction [5]. The diagnosis relies on clinical manifestations, with the most frequently cited profile composed of the triad of abnormal uterine bleeding (50%), secondary dysmenorrhea (30%), and an enlarged, tender uterus [6], sometimes accompanied with subfertility [7,8].

The transabdominal ultrasound (TAS) characteristics include a honeycomb pattern, sonolucent 5–7 mm spaces, myometrial cysts, wide posterior wall, eccentric endometrial cavity, diffuse uterine enlargement but with no leiomyomas, and diminished uterine echogenicity [9–14]. However, the preoperative diagnosis of adenomyosis cannot be reliably differentiated from that of leiomyoma on TAS [9]. The transvaginal ultrasound (TVS) findings include uterine enlargement not explainable by the presence of leiomyomas, asymmetry (asymmetrical thickening of the anterior or posterior uterine wall), lack of contour abnormality or mass effect, retroversion, globular or spherical shape, and uterosacral ligament nodularity for uterine body examination; focal honeycomb, scattered, irregular, cystic, anechoic lacunae, heterogeneous increased or decreased myometrial echoes, mottled texture, lesions surrounded by anechoic shadows 1–3 mm in diameter, hyperechogenic or hypoechogenic striations, indistinct margins, nonencapsulated myometrial lesion, and myometrial linear striation in the myometrium examination; and a shaggy endometrial stripe, blurred endometrial–myometrial border, and thickened or distorted endometrium in the endometrial examination. Although these features are commonly imaged in women with adenomyosis, there is no general agreement on which diagnostic criteria have the highest diagnostic accuracy. According to Kepkep et al, the presence of subendometrial echogenic linear striations might be the most specific finding for the diagnosis of uterine adenomyosis [10]. The presence of subendometrial echogenic linear striations, a heterogeneous myometrial echotexture, and myometrial anterior–posterior asymmetry on TVS supports the diagnosis of adenomyosis [11]. A recent systematic review and meta-analysis showed that TVS is a moderately accurate test for the diagnosis of adenomyosis, but as a preoperative test with ease of use, cost effectiveness, and wide availability, it likely represents the most practical diagnostic test for adenomyosis [9].

Magnetic resonance imaging (MRI) findings [12,13,15,16] may be more helpful than TVS in distinguishing adenomyosis from a leiomyoma and more effective for both diffuse and focal adenomyosis, with sensitivity and specificity that are comparable with or even better than those of ultrasound, as it depicts contrasts between low-intensity lesions and surrounding tissues; however, it is more expensive. The characteristic findings of MRI in uterine adenomyosis are a minimum width at the junction zone of 5 or 12 mm, a focal and uneven width of the junction zone, low intensity of the junction zone, high-intensity spots scattered within the junction zone, and indistinct junction zone margins for the myometrial examination. From the histological perspective, areas of low-signal intensity correspond to smooth muscle hyperplasia, and bright foci on T2-weighted images correspond to islands of ectopic endometrial tissue and cystic dilatation of glands [16]. When menstrual hemorrhage occurs within these ectopic endometrial tissues, signal intensity on T1-weighted images may become high.

Challenge in the management of adenomyosis

Because the definite diagnosis of adenomyosis is based on a histological examination, usually on a hysterectomy specimen [17], adenomyosis is a neglected diagnosis [18]. Hysterectomy remains a most important therapeutic option for women with symptomatic adenomyosis [9], although medical management can be effective [19–26], similar to the management of uterine fibroids (myoma) [26–34]. However, its effect is often transient and it is frequently used with a preoperative adjuvant therapy or sometimes a postoperative therapy [35–37]. Rapid regrowth of adenomyosis and relapse of symptoms and signs always occurs after the treatment is stopped [38]. Therefore, other alternative strategies might be needed. Surgery might be one of the most familiar of these uterine-sparing procedures [38–45]. The following sections summarize recently advanced technologies and the outcomes of patients with adenomyosis and/or adenomyoma after uterine-sparing surgical treatment.

Surgical treatment

Compared with the well-accepted gonadotropin-releasing hormone agonist as a choice in the management of symptomatic women with extensive adenomyosis who want to preserve their uterus, conservative uterine-sparing surgery is seldom considered as playing a role in managing these patients, partly because there is difficulty in selecting a good candidate to undergo this surgical approach, and partly because it is not easy to determine the extent of the adenomyosis involved and how much of the tumor should be removed. Another consideration, based on the characteristics of the disease itself (heterotopic endometrial glands and stroma widely spreading and scattering throughout the myometrium, with adenomyosis foci frequently mixing with the surrounding normal myometrium), was that it is nearly impossible to complete resection of uterine adenomyosis using conservative uterine-sparing surgery. In addition, the uterine cavity is frequently entered during the operation. One study showed that nearly 30% of patients needed careful repair of the uterine cavity during the tumor resection of adenomyosis [38]. Otherwise, it is more difficult to repair the uterine defect after conservative uterine-sparing surgery for adenomyosis than for leiomyoma [46], resulting in poor healing of the uterine defect or weakness of the uterine scar after surgery. The uterine scars after conservative uterine-sparing surgery of adenomyosis might not return to normal tensile strength, partly because the defect may contain and conceal more adenomyotic foci and partly because repair is neither adequate nor secure enough. The decreased tensile strength of the uterus might also fail to be competent enough for future pregnancy, because the ability of the uterus to enlarge and distend along with an advanced-stage pregnancy was impaired, resulting in a significantly increased risk of a ruptured uterus during pregnancy and labor [47]. Because a similar risk has been reported in many patients with a history of myomectomy [48,49], it is reasonable to expect a much greater possibility of uterine rupture in these women after conservative uterine-sparing surgery for adenomyosis when they are pregnant. Besides the aforementioned considerations, there are many potential risks or possible sequelae following uterine-sparing surgery, such as pelvic adhesion, uterine deformities, intrauterine adhesions, and reduced uterine capacity. Therefore, the decision to use conservative uterine-sparing surgery in the management of women with extensive adenomyosis should be taken carefully.

Classification of conservative uterine-sparing surgery for adenomyosis

An excellent review addressing *uterus-sparing operative treatment for adenomyosis* was published online in November 2013 [3]. A