Case Report

Adenomyotic Cyst in a 25-Year-Old Woman: Case Report

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ABSTRACT

Adenomyotic cysts are uncommon findings, usually in the context of diffuse adenomyosis and <5 mm in diameter. Herein we report a 4.5-cm adenomyotic cyst in a 25-year-old nulliparous woman with severe dysmenorrhea and pelvic pain. Transvaginal ultrasonography and magnetic resonance imaging revealed a well-circumscribed hypoechogenic mass in the posterior uterine wall, well separated from the uterine cavity. Pathologic analysis demonstrated that the cyst was lined with endometrial epithelium and stroma and was surrounded by smooth muscle hyperplasia. In the literature, we found 30 reports of cysts with similar characteristics. Because this cyst has not been clearly defined, it has been called by various names including adenomyotic cyst, cystic adenomyosis, and cystic adenomyoma. We believe this lesion should not be called an adenomyoma, but is more correctly called an adenomyotic cyst or, depending on age at onset, a juvenile adenomyotic cyst. Journal of Minimally Invasive Gynecology (2013) 20, 894–898 © 2013 AAGL. All rights reserved.

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DISCUSS

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Adenomyosis is the presence of endometrial glands and stroma in the context of the myometrium, with adjacent smooth muscle hyperplasia. It may be diffuse or cystic. Diffuse adenomyosis occurs more commonly, with clinical findings of cystic spaces filled with blood, usually <5 mm in greatest diameter [1]. Cysts ≥ 1 cm in diameter are uncommon; to date, only about 30 cases have been reported. Large adenomyotic cysts are lined with eutopic functional endometrium-like tissue and are characterized by cyclic changes with epithelial exfoliation and hemorrhagic infarction of adjacent smooth muscle.

Adenomyosis usually affects women aged <30 years who have never undergone any gynecologic surgical inter-

vention. Symptoms include severe and worsening secondary dysmenorrhea associated with menorrhagia and pelvic pain.

Case Report

A 25-year-old nulliparous woman came to our institution because of severe and worsening dysmenorrhea and abdominal cramping. Her first menses occurred when she was aged 12 years, and her menstrual cycle was regular (25 days). Dysmenorrhea began shortly after menarche; however, the pain was sufficiently relieved with use of analgesics (nimusulide or paracetamol).

In the past year, the pain had become more severe and refractory to any medical treatment. Typically, the pain began 1 week before menses and continued in the week afterward. The patient reported that the pain was more intense on the first day of each menstrual cycle. According to a visual analog scale ranging from 0 (no pain) to 10 (extreme pain), the severity of dysmenorrhea was 9. The patient also had menorrhagia but no dyspareunia or dyschezia.

Pelvic examination revealed a normal vagina, vulva, and adnexae and an enlarged retroflexed uterus. Laboratory
findings including complete blood cell count, complete metabolic panel, liver function tests, pancreatic enzyme levels, urinalysis, and erythrocyte sedimentation rate were within normal limits. CA 15.3, carcinoembryonic antigen, and β-human chorionic gonadotropin levels were normal (12.80 U/mL, 1.35 ng/mL, and 1.20 mU/mL, respectively), whereas CA 125 levels were slightly elevated (38.00 U/mL), and CA 19.9 level was high (88.19 U/mL). Transvaginal ultrasound, performed immediately before surgery, showed a well-circumscribed hypoechogenic mass, similar to an endometrioma, in the posterior uterine wall, well separated from the uterine cavity. The cyst cavity measured 4.5 × 2.4 cm in diameter and was surrounded by a thick capsule, for a total diameter of 5.0 × 3.4 cm (Fig. 1). Transvaginal ultrasound performed during the luteal (secretory) phase of the menstrual cycle confirmed no evidence of communication between the lesion and the endometrium. Both ovaries appeared normal. Pelvic magnetic resonance imaging disclosed an retroflexed enlarged uterus (7.2 × 8.2 × 8.7 cm) and a rounded well-defined intramyometrial mass in the posterior wall of the uterine corpus. The mass measured 3.2 × 4.5 × 4.1 cm, and was slightly hyperintense on T1- and T2-weighted images, with an internal fluid-fluid level, i.e., layering of simple and hemorrhagic or proteinaceous fluids (Fig. 2). Small masses in both ovaries (right, 12 mm; left, 7 mm) with the same findings as those of the intramyometrial mass were noted.

Operative laparoscopy was performed after 3 months, on day 24 of the menstrual cycle, with the patient under general anesthesia. With the patient in a lithotomy position (±20-degree Trendelenburg position), pneumoperitoneum was induced using a Veress needle, and trocars were inserted. At laparoscopic observation, the posterior wall of the uterine corpus demonstrated an altered profile. After the external layers of the mass were incised, a chocolate-colored fluid was drained. During the surgical procedure, intrauterine methylene blue injections demonstrated no communication between the endometrial cavity and the cyst, and both fallopian tubes were patent.

The surgical procedure consisted of radical excision of the adenomyosis, leaving a 1-cm margin of tissue above the endometrium and a 1-cm margin of tissue below the serosal surface, with subsequent reconstruction of the uterus. The enlarged uterus was bisected using a monopolar hook, from the serosal surface of the fundus, in the midline and in the sagittal plane, down through the adenomyosis. Thus, the entire extent of the adenomyosis was clearly visible, with the crucial landmarks of the endometrium and the serosal surface always in clear view. The adenomyotic tissues were grasped and excised from the surrounding myometrium, leaving 1 cm of myometrial tissue from the serosa above and the endometrium below. Thereafter, the normal myometrium was closed using interrupted absorbable suture (Fig. 4). Small foci of endometriosis were detected in the right ovary and the pouch of Douglas, and were treated using bipolar electrocautery. Operative time was 70 minutes, and blood loss was <20 mL. The postoperative course was normal, and the patient was discharged 2 days after the intervention.

Postoperatively, a gonadotropin-releasing hormone agonist was administered for 3 months. At menstruation, the patient had no dysmenorrhea. At macroscopic examination, the lesion measured 4.5 cm in diameter. Biopsy specimens demonstrated endometrial epithelium and stroma lining the inner cyst wall, bordered by a region of myometrial hyperplasia. Hemorrhage and hemosiderin-laden macrophages were seen in association with endometrial sloughing (Fig. 3).
Discussion

Cystic structures within the uterine cavity are uncommon [2,3]. Uterine cysts are classified into 2 main groups: congenital and acquired. Acquired cysts include cystic degeneration of uterine leiomyoma, cystic adenomyosis, and serosal cysts. Congenital cysts derive from müllerian (paramesonephric) duct anomalies such as a non-communicating rudimentary horn and a unicornuate uterus or can be represented by wolffian duct (mesonephric) and müllerian duct cysts [4].

Large adenomyotic cysts (≥1 cm in diameter) are rare. Although any site in the uterus may be involved, the posterior wall is most often affected [5].

There is no agreement about the definition of adenomyotic lesions. Several terms have been used to define these large cysts including adenomyotic cyst [6–8], cystic adenomyoma [9–11], and cystic adenomyosis [12,13]. According to Takeuchi et al [14], adenomyoma is a neoplastic tumor classified as a benign mixed epithelial-nonepithelial tumor. The designation of adenomyoma should be reserved for polypoid lesions in which the stromal component is almost exclusively composed of smooth muscle. The term adenomyoma is not recommended to define a solitary myometrial nodule of adenomyosis [15].

Furthermore, there is doubt about the nature of such rare voluminous cysts in young women. Considering patient age at symptom occurrence, some authors have classified these lesions into 2 categories: the adult and the juvenile forms. The adult form seems to result from trauma at the endometrium-myometrium interface, e.g., after uterine instrumentation [16]. The juvenile form is considered a congenital disease that develops from duplication and persistence of ductal müllerian tissue in a critical area, close to the root of the round ligament, possibly related to gubernaculum dysfunction [17]. Other authors [3,14] have described both the juvenile and adult forms as of acquired origin, probably related to a cystic variant of adenomyosis.

At histologic analysis, adenomyotic cysts are cavities lined by endometrial epithelium [18]. The stroma below the epithelium is thin throughout the cyst and contains red blood cells and hemosiderin-laden macrophages. The stromal cells are morphologically similar to those of the endometrium, as in endometriosis.

In conclusion, taking into consideration the relevant literature, the clinical findings, and the pathologic features, we consider the large cyst in our patient to be an acquired adenomyotic cyst. That the cyst was acquired is suggested by onset of dysmenorrhea long after menarche, absence of other

![Fig. 3](https://example.com)

Histologic findings of adenomyotic cyst. (A) Single layer of endometrial epithelium with underlying endometrial-type stroma (H&E × 20). (B) Adjacent myometrium shows an area of extensive hemorrhage (H&E × 20). (C) Stromal tissue is stained positively for CD68, proving the presence of macrophages × 40. (D) Endometrium-like tissue lining the cyst is stained positively for human chorionic gonadotropin × 40.
associated congenital defects, absence of any communication with the uterine cavity, and location far from the round ligament.

The presence of endometrium surrounding the internal cavity, composed of a single columnar layer accompanied by endometrial stroma, hemorrhage, and hemosiderin-laden macrophages, is a clear characteristic of adenomyosis. We believe this lesion should not be called an adenomyoma because it did not have a relevant smooth muscle component; rather, it is more correctly called an adenomyotic cyst. Because of the age at onset, it could more exactly be called a juvenile adenomyotic cyst.

Further case reports are required to better understand the pathogenesis and clinical characteristics of this rare disorder.

References