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Case Report

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# LARGE ADENOMYOTIC CYST IN A 35-YEAR-OLD WOMAN

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## ABSTRACT

Adenomyotic cyst is a rare form of adenomyosis caused by extensive menstrual bleeding into the ectopic endometrium. Exceptionally, they can show massive growth resulting in what is called large adenomyotic cyst. We report a case of a 35-year-old woman presented with chronic pelvic pain and severe dysmenorrhea appeared five months after her last caesarean section. Abdominopelvic ultrasound showed a large cystic lesion in the posterior uterine wall suggestive of an adenomyotic cyst which was confirmed after surgical enucleation.

**KEYWORDS:** Adenomyosis; Adenomyotic cyst; Ultrasound; Surgery.

## INTRODUCTION

Adenomyotic cyst (AC) is an extremely rare form of adenomyosis caused by extensive menstrual bleeding into the ectopic endometrium. Usually, the cyst is less than 10 mm in diameter and only few cases with large diameter have been documented to date.[1] We present a case of large adenomyotic cyst in a 35-year-old woman with the goal of increasing awareness of this uncommon condition among clinicians for early diagnosis and appropriate treatment.

## CASE REPORT

A 35-year-old multiparous woman presented in our department with chronic pelvic pain and severe dysmenorrhea appeared 7 months before. She had two previous caesarean sections and the last one being 1 year ago. Her menstrual cycle was regular. Clinical examination revealed a large pelvic mass extending up to the umbilicus. The mass was firm in consistency with restricted mobility and its lower border could not be reached. Abdominopelvic ultrasound showed a well-circumscribed cystic lesion, measuring 20 cm × 12 cm, in the posterior uterine wall. The mass had no communication with the endometrium and both ovaries appeared normal. Routine laboratory tests and serum cancer antigen 125 levels were within normal limits. The presumptive diagnosis was an adenomyotic uterine cyst. Under general anesthesia, pfannenstiel incision was realized; after opening the parietal peritoneum, a giant bulging cystic mass in the posterior wall of the uterus reaching the uterine serosa was seen. Both ovaries and fallopian tubes were grossly normal. A transverse incision was made in the uterine serosa followed by enucleation of the cystic lesion which was filled with chocolate-colored fluid (Figure 1). Visible vessels were coagulated and no communication between the cyst and endometrial cavity was seen. The myometrium was closed using an interrupted absorbable suture. Total operating time was 45 minutes and the estimated blood loss was less than 50 ml. Histopathologic examination objectified a cystic structure lined by endometrial epithelium and surrounded by a region of myometrial hyperplasia confirming the diagnostic of an adenomyotic cyst. The postoperative course was uneventful and no recurrence was seen with a follow up period of 3 years.

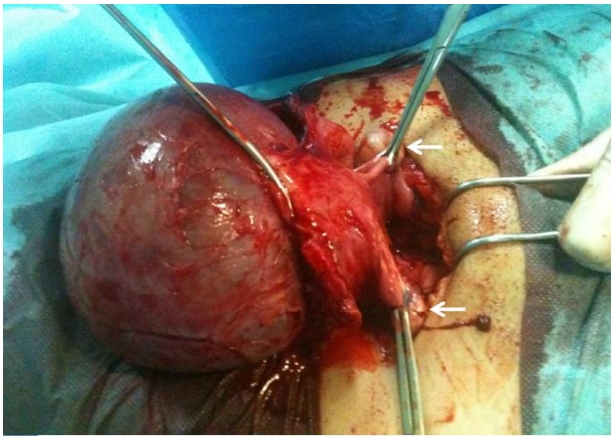

**Figure 1: Peroperative view of the adenomyotic cyst, note the normal appearance of ovaries (white arrows).**

## DISCUSSION

Adenomyotic cyst was first described by Cullen in 1908 in five hysterectomy specimens.[2] It is a rare variant of adenomyosis and develops when there is bleeding into ectopic islands of endometrial glandular tissue surrounded by myometrium. Repeated hemorrhage

during menstruation is considered to be a cause of extensive cyst formation. Although any site in the uterus may be involved by this entity, the posterior wall is most often affected, as was in our case.[3] Considering patient's age at symptom development, ACs are classified into juvenile and adult form. The adult form, which is usually found in multiparous women over age 30, seems to develop when the endometrium is sutured into the myometrium during uterine surgery (after cesarean section, myomectomy or curettage).[4] 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 a gubernaculum dysfunction. In the present case, we classified the large AC into adult form based on history of two caesarean sections, patient's age at symptom development, absence of other associated congenital defects and location of the cystic lesion far from the round ligament. In vast majority of reported cases, diagnosis of AC was only made after surgical exploration given its rarity and its appearance which can mimic other uterine tumors, such as leiomyomas with cystic degeneration, or even cystic ovarian tumors. Other differential diagnoses include congenital uterine cysts, intramyometrial hydrosalpinx and echinococcal cyst. In 2015, Pontrelli et al. described a case of giant cystic adenomyoma erroneously diagnosed as a large cornual hematometra in a bicornuate uterus.[5] The primary condition for the diagnosis of AC is clinical awareness of the possibility of such lesions. This is particularly relevant in young women presenting with severe dysmenorrhea and pelvic pain that does not respond to common analgesics. Imaging techniques are critical in differential diagnosis of ACs and are useful to choose the appropriate management by also taking into account the size and the localization of the cyst and patient's age. Ultrasonographic features of ACs have been described and are often nonspecific, including both homogeneous and heterogeneous cystic lesions. However, magnetic resonance imaging (MRI) can be crucial for making a correct preoperative diagnosis. MRI features include a complex cystic lesion located within the myometrium, with hyperintense T1-Weighted and intermediate to hyperintense T2-Weighted signal contents. Another important characteristic is the presence of surrounding T2-hypointense tissue compatible with reactive myometrial hypertrophy.[6] In this case, two factors led us to consider preoperatively AC as the cause of the pelvic pain and dysmenorrhea. Firstly, the patient had never experienced such symptoms before the last cesarean section, but after the event, she reported progressive pelvic pain with the maximum during her menstrual cycle. Secondly, sonographic appearance of the lesion was of an endometrioma in the posterior wall of the uterus, the cyst had no communication with the endometrium and both ovaries appeared normal. We believe that this is the first case of a large AC following cesarean section in which the preoperative diagnosis was based only on clinical and sonographic features. Surgical enucleation is the gold standard treatment of these benign lesions. AC is more clearly delineated from the normal myometrium than uterine adenomyosis, but less than uterine myomas. In our patient, laparotomy was preferred compared to laparoscopy given surgical history of two cesarean sections and the myometrium was closed with interrupted suture, after enucleation of the cystic mass, similar to the procedure adopted after myomectomy. Operative hysteroscopy may be used for the submucosal or intramural adenomyotic cyst but its effectiveness has not been properly investigated.[5] Medical management with levonorgestrel intrauterine system is an effective method in alleviating dysmenorrhea in women with adenomyosis, but there are no reports of its use in women with ACs.[7]

## CONCLUSION

This case suggests that adenomyotic cyst must be considered in women of reproductive age presenting with severe dysmenorrhea and persistent pelvic pain after uterine surgery. Work-up must include both imaging and intraoperative findings. Conservative surgery is recommended to excise the lesion completely and preserve fertility.

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