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Cystic adenomyosis spreading into subserosal-peduncolated myoma: How to explain it?

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ABSTRACT

INTRODUCTION: Cystic adenomyosis is a rare variant of adenomyosis characterized by well- circumscribed cavitated endometrial gland and stroma located within the myometrium. The cysts usually measure ≥ 1 cm in diameter, contain a "chocolate-colored" fluid and do not open into the overlaying endometrium.
CASE PRESENTATION: We present a case of a peduncolated-subserosal cystic adenomyoma, namely cystic adenomyosis, correlated with pelvic MR imaging, laparoscopic surgery technique and histopathology findings.
CONCLUSIONS: In this case, the peculiar growth pattern of cystic adenomyosis in a myoma represents a singular condition rarely reported in the medical literature. We therefore support the pathogenetic theory that the disease might have been caused by direct proliferation of endometrial cells within a peduncolated- subserosal myoma.

1. Introduction

Adenomyosis is a benign condition characterized by myometrial spreading of the endometrium [1], usually involving the basal layer. Clinically, the disease may cause severe, secondary dysmenorrhea associated with menorrhagia and chronic pelvic pain [2]. Adenomyosis commonly appears within the myometrium as clusters of small cystic spaces filled with blood [3] , that have rarely a diameter greater than 5 mm [4]. In rare cases, the lesion may be seen as a single cyst [5], with a diameter ≥ 1 cm, filled with a chocolate-brown-coloured fluid and namely cystic adenomyosis [2]. We report a rare case of cystic adenomyosis found in a subserosal pedunculated myoma, that is to say cystic adenomyoma. Etiopathogenetic mechanisms of the condition are also speculated.

2. Presentation of case

A 39-year-old woman, with unremarkable past medical history, was referred for gynaecologic consultation due to severe, recurrent dysmenorrhea associated with chronic pelvic pain unresponsive to medical therapy. Trans-vaginal ultrasound performed using a 6.5–7.5 MHz probe showed two peduncolated-subserosal ovoid masses located on the anterolateral and posterior right side of the uterine wall, respectively. A presumptive diagnosis of myomas was prompted. Increased serum CA125 level was documented (122.4 U/m, range 0–35 U/ml). Pelvic MR imaging performed using a 1.5 Tesla equipment, confirmed the ultrasound diagnosis. In addition, the high-intense anterolateral mass of 6 cm showed two hypointense (about 2 cm each one), contrast-enhanced central area on T2-w (weighted) images (Fig. 1). Therefore, MRI diagnosis was subserosal myomas with marked central necrosis in one (the anterolateral one). Differential diagnosis was posed with adenomyosis. Laparoscopic surgery was undertaken with radical resection of the peduncolated- subserosal masses.

The laparoscope was inserted from a 10 mm umbilical port. Ancillary 10 mm lateral port was then placed to the left while two others 5 mm ports were inserted centrally and to the right under direct laparoscopic guidance (Fig. 2). At the time of the *morcellament* of the "core" of the anterolateral myoma, a "chocolate colored" fluid spillage was noted (Fig. 3; Video S1). Histological examination showed ectopic and cystic endometrial glands and stroma within the myoma, a finding consistent with a peduncolated cystic adenomyoma. No malignant features were identified (Fig. 4).

3. Discussion

The adenomyotic cyst shows clinical characteristics similar to those of common adenomyosis, which is usually found in

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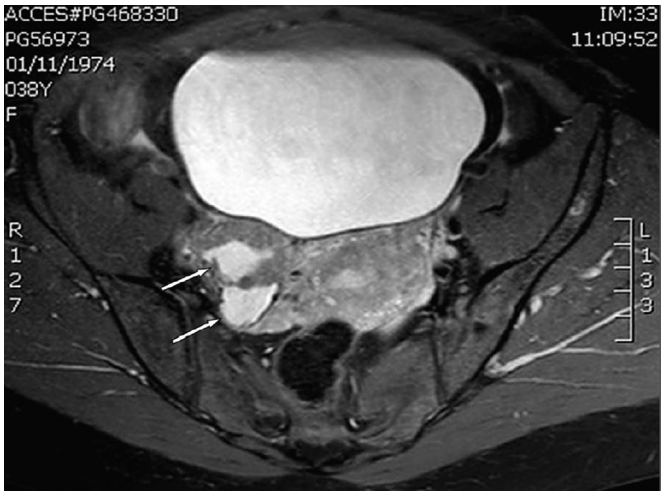

Fig. 1. T2-weighted contrast- enhancement MR imaging showing the two hypointense signal inside the antero-lateral and right-sided myoma (white arrows).

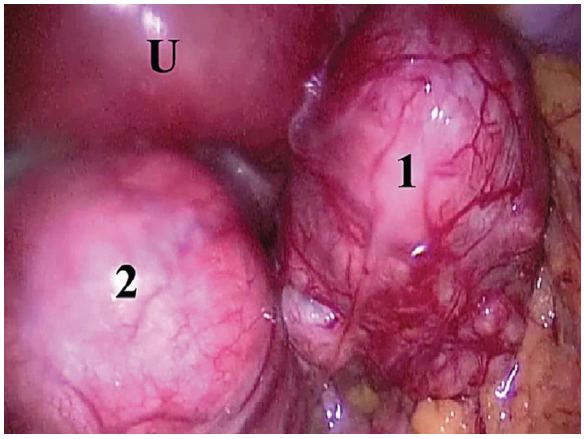

Fig. 2. Laparoscopic finding of the uterus (U) with the antero-lateral [1] and the posterior myoma [2].

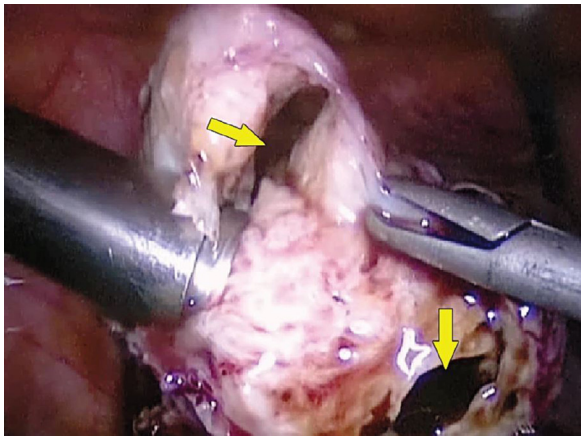

Fig. 3. Laparoscopic finding at the time of *morcellament* of the "core" of the antero-lateral myoma. The spillage of "*chocolate colored*" fluid is clearly seen.

multiparous women over the age of 30 [6]. It is considered an extremely rare variant of adenomyosis characterized by the presence of a hemorrhagic cyst resulting from menstrual bleeding in the ectopic endometrial glands [7]. Ho et al. [8] have reported a rare manifestation of adenomyosis - the adenomyotic cyst - in a 16 -year-old girl with chronic pelvic pain.

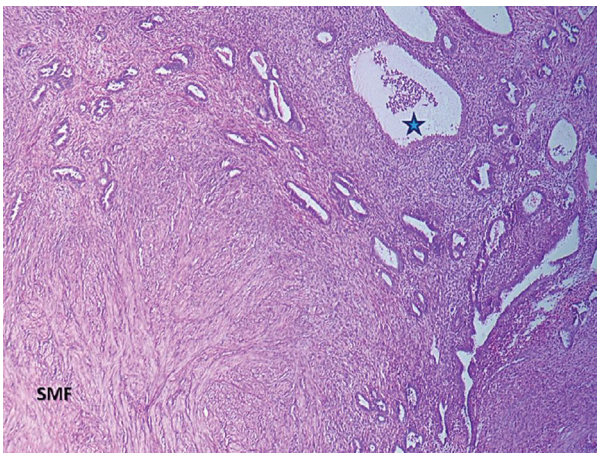

Fig. 4. Pathology. Diagnostic patterns of adenomyosis are demonstrated: endometrial glands and stroma intermixed with smooth muscular fibers (left-hand and upper side). Smooth muscle fibers (SMF) are more prominent with no glandular structures (left and bottom side). The glands are enlarged with cellular debris in the lumen (blue star, right-hand side). This histologic findings are consistent with a diagnosis of cystic adenomyosis.

Adenomyomas are benign uterine tumors of unknown etiology that mainly affects perimenopausal women [9,10] whilst the juvenile form has been reported only in ten cases [11,12]. Of note, cystic degeneration is often seen in leiomyomas, but rarely reported in adenomyomas [11] .

Diagnostic MRI cluster of cystic adenomyosis rely on the detection of a complex cystic lesion that is usually located within the myometrium, with hyperintense T1-w and intermediate to hyperintense T2-w signal contents suggesting hemorrhagic and/or proteinaceous products. In addition, presence of surrounding T2-hypointense tissue, is indicative of reactive myometrial hypertrophy and/or thin rim cystic wall may showhypointense T1-w and T2-w signal, suggestive of presence of hemosiderin due to endometrial sloughing [10,13,14].

The exact mechanisms of how adenomyosis develop are still unknown, but different theories have been proposed. The foremost hypothesis suggests an origin from the deep part of the endometrium that would invaginate between the bundles of smooth muscle fibers of the myometrium itself. The process of invagination and myometrial spreading may be facilitated by the non-cyclic, anti-apoptotic activity of the basalis associated with relative hyper-estrogenic states [1] . Cullen [15] reported that adenomyosis can bulge at the outer peritoneal surface, forming a sub-peritoneal adenomyoma. This lesion is prone to becoming cystic with cavities being filled with "chocolate-colored" content [16]. Nonetheless, difficulties are encountered to support this hypothesis when the ectopic tissue is seen involving areas distant from the normal endometrium [17] or in cases with no previous gynecologic surgery. La Fianza et al. [18] described a large isolated adenomyosis with a pedunculated appearance and have explained it as the possible presence of ectopic endometrial islands in the outer portion of the myometrium. The exophytic growth of the lesion or the cystic dilatation of the glands could have been due to blood pooling, with progression outside the uterine wall. Considering the common embryological origin from the Müllerian ducts of the endometrium and the subjacent myometrium [1], a direct proliferation of metaplastic myometrial cells of endometrial tissue may be a possible pathogenetic mechanism of cystic adenomyosis. Pistofidis et al. have recently reported that in sixty-eight women, who underwent laparoscopic treatment of adenomyosis, only 4.5% showed cystic features [19].

4. Conclusion

Pedunculated cystic adenomyoma, as the one described, represents a gynaecologic condition that has been seldom reported in the medical literature. This rare entity might have been the result of metaplastic myometrial cells differentiated in ectopic endometrium. Clinical diagnosis may be challenging, but contrast hysterosalpingography (HSG) and MRI support ultrasound diagnosis and help in planning the best surgical approach.

Key learning point

A support to the pathogenic theory about the origin of adenomyosis.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at http://dx.doi.org/10.1016/j.ijscr.2015.01.005.

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