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Case Report

Myometrial-Adenomatoid Tumor with Unusual Diffuse Myometrial Involvement and Co-existing Endometriosis-Case Report and Literature Review

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Abstract

Adenomatoid tumors are rare, benign neoplasms that primarily affects the genital and urinary tracts. Only a few cases of myometrial adenomatoid tumors treated laparoscopically or in co-existence with endometriosis have been reported. Here we report a case of laparoscopically resected myometrial adenomatoid tumor with co-existing endometriosis and unusual histological features. A 30-year-old nulliparous woman with subfertility and ultrasound diagnosed uterine fibroid underwent laparoscopic excision of uterine mass and excision of endometriosis. Intra-op enucleation of the uterine tumor was challenging due to an ill-defined border between the lesion and surrounding normal myometrium. Histological features were misleading due to irregular proliferation of smooth muscle cells. The myometrial adenomatoid tumor displayed unusual features of large size, diffuse myometrial involvement with solid proliferation of mesothelial cells and possible serosal involvement, explaining the diagnostic and operative challenge.

Keywords: *Myometrial Adenomatoid Tumor; Adenomatoid Tumors; Leiomyomas; Endometriosis; Benign Tumor*

Background

Adenomatoid tumors (AT) are rare benign tumors of mesothelial origin. They predominantly occur in the genital tract in both males and females [1]. Occasionally, AT have been reported in other sites such as, mesentery, mediastinum, pleural cavity, adrenal gland, pancreas, heart, and mediastinal lymph node. In females, these tumors are typically found in the subserosa of the cornual myometrium, adnexa, or in proximity to the fallopian tubes [2]. Uterine lesions are typically solitary, ill-defined and occupy more than 50% of the myometrium [3].

Microscopically, adenomatoid tumors are characterized by the presence of inter-anastomosing pseudo-glands or pseudo-vascular spaces [4]. These structures mimic glandular formations and can resemble vascular structures, respectively. In some cases, adenomatoid tumors may also exhibit the prominence of smooth-muscle component, histologically mimicking leiomyoma. Hence, this sub-type has been termed 'leiomyo-adenomatoid tumor' (LMAT) [4]. In LMAT sub-group of AT, the adenomatoid component may mimic a malignant tumor due to irregular pseudo infiltration with tubular formations [5].

There is a sparse amount of published literature review few cases of myometrial adenomatoid tumor with co-existing endometriosis [6,7]. In this report, we present a case of uterine myometrial adenomatoid tumor with unusual features co-existing with endometriosis and infertility posing a diagnosis challenge.

Case Report

A 30-year-old nulliparous woman who was referred from a fertility center due to the incidental finding of a solitary 10 cm uterine fibroid during evaluation for fertility treatment. The patient was asymptomatic, undergoing *in vitro* fertilization (IVF) treatment, and awaiting embryo transfer. Pelvic Ultrasound imaging was performed, revealing a 10 cm fundal sub-serosal fibroid with no suspicious features. Subsequently, during laparoscopy, a solitary 10 cm type 6 fundal fibroid (Figure 1) was identified, accompanied by stage II endometriosis, according to AAGL Endometriosis Classification, involving the left uterosacral ligament, right pelvic side wall and right ovary, and recto-vaginal (RV) septum (Figure 2-4). The uterine fundus was injected with diluted vasopressin using a laparoscopic needle, and an incision was made with Thunderbeat™ (Olympus). Laparoscopic excision of the clinically presumed fibroid was challenging due to the ill-defined borders between the tumor and normal myometrium. After resection of the tumor, uterine wall was sutured in layers using V-lock 0 suture. The recto-vaginal nodule was excised with cold scissors and endometriotic deposits on ovary had plasma targeted treatment (Helica™). The total operative time was 90 minutes. The patient experienced an uneventful postoperative recovery and was discharged home on the same day.

Initially, the histopathological examination revealed findings consistent with benign leiomyomata, exhibiting prominent blood vessels, lymphoid aggregates, and proliferation of small solid nests and tubules with bland nuclei and abundant eosinophilic cytoplasm. To further characterize the tumor, immunohistochemistry was conducted, which demonstrated positive staining with epithelial marker AE1/3, Calretinin, and weakly positive staining with PAX8. Further pathological review revealed the unusual features of the adenomatoid tumor. The unusual aspects of the tumor included its large size, diffuse involvement of the myometrium, and solid proliferation of mesothelial cells with serosal involvement.

Laparoscopic findings

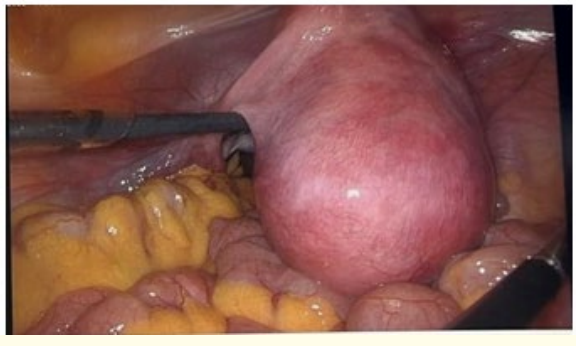

Figure 1: *10 cm fundal fibroid. Figo type 6.*

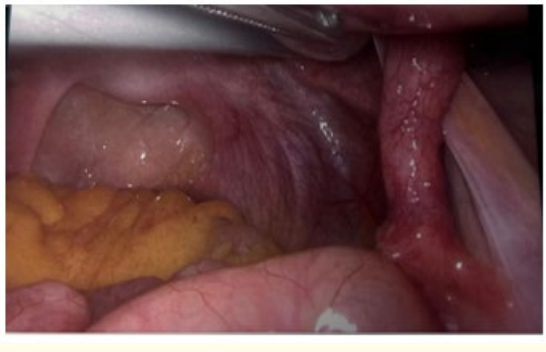

Figure 2: *Right pelvic side wall endometriotic nodule.*

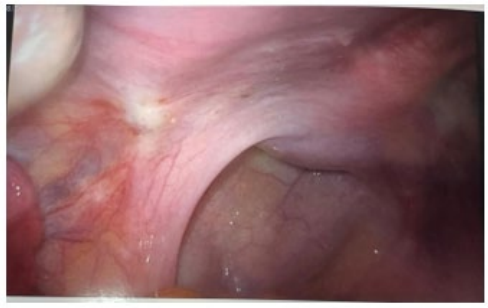

Figure 3: *Left uterosacral endometriotic nodule.*

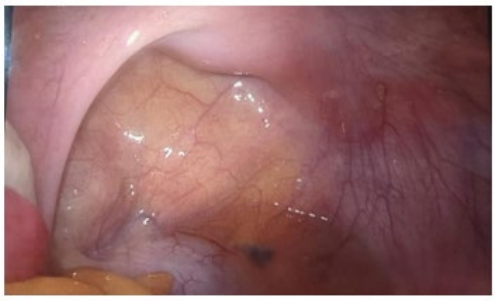

Figure 4: *Rectovaginal endometriotic nodule.*

Discussion

A leiomyoma-adenomatoid tumor was first described by Epstein in 1992, as a morphological variant of adenomatoid tumors with a prominent smooth muscle component [8]. They are rare benign neoplasms which are incidentally detected during histopathologic evaluation of routine myomectomy or hysterectomy specimens for fibroids. Typical morphological features of leiomyoma-adenomatoid tumors are solitary, small (< 4 cm), and solid tumors grossly mimicking leiomyomata. Rarely, they can be multifocal, large (> 10 cm), diffuse or cystic [4].

The incidence of leiomyoma-adenomatoid tumors captured in literature for hysterectomy done for other reasons is estimated to be approximately 1 - 1.2% [9]. They typically occur in women of late reproductive age (average age is 45). Typically, it occurs in the posterior wall of the uterus, and > 80% are solitary lesions. Histopathological diagnosis of AT is generally made with hematoxylin-eosin stating. Also, AT are immunohistochemically positive for mesothelin, thrombomodulin, and podoplanin. Lerias., *et al*. had described a series of four cases of unusual adenomatoid uterine tumour with diffuse myometrial involvement, and we believe our case has similar unusual features [3]. The exact pathogenesis of AT is still unknown. Nogales., *et al*. suggested that AT may arise from mesothelial cells. This hypothesis may explain the co-existence of endometriosis in some cases [1].

To date, no laparoscopic surgical consensus has been established for the management of adenomatoid tumors. Much of the available literature on AT have focused on the pathological features of the tumor with scarce data available on clinical aspects and surgical management of AT. As Sakurai described, excision of AT needs careful evaluation and specific skills as borders are ill-defined between AT and myometrium [6].

Conclusion

In conclusion, adenomatoid tumours are rare, benign neoplasms primarily affecting the genital and urinary tracts. Accurate diagnosis is crucial to differentiate them from other lesions such as leiomyomas, adenomyosis, or other malignant or metastatic lesions. While most cases can be managed conservatively, surgical excision may be necessary in selected cases. Future research should focus clinical aspects, correlation with endometriosis and surgical approaches of the adenomatoid tumour.

Declaration of Interest

None.

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