

Analysis of diagnosis and treatment of complicated cervical severe adhesion atresia after removal of endometrial stromal nodule

A case report

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Abstract

Rationale: Endometrial stromal nodule (ESN) is a rare benign endometrial stroma tumor. Experiences are helpful for avoiding and treating similar postoperative complications (cervical adhesions and atresia).

Patient concerns: When appearing in the cervical, this tumor can easily lead to complications after the surgical resection. The diagnosis and postsurgery complication of a young woman's ESN was reported here.

Diagnoses: The postoperative pathological diagnosis was ESN.

Interventions: A 29-year-old young woman was diagnosed and treated for ESN in cervical parts with postsurgery complications of cervical complex adhesion atresia.

Outcomes: The complication was complex cervix adhesion atresia with very special imaging performance—the cervix and the palace imaged as “Twisted and Angled Staircase.” This particular cervix adhesion was challenging for operation. We achieved a successful treatment through the carefully designed surgical procedure including the application of hysteroscopy and laparoscopy.

Lessons: The lower uterine segment and cervix should be paid attention during suturing in this situation. Close and positive follow-ups should be planned after the endometrial stromal resection. The reconstruction of the tunnel is a solution for the problem of menstruation.

Abbreviation: ESN = endometrial stromal nodule.

Keywords: cervical adhesion atresia, endometrial stromal nodules

1. Introduction

Endometrial stromal tumor is a rare gynecological tumor, accounting for 2% of all uterine tumors.^[1] With development of molecular genetics technologies, the World Health Organization divided endometrial stromal tumors into 4 types: endometrial stromal nodule (ESN), low-grade endometrial stromal sarcoma, high-grade endometrial stromal sarcoma, and undifferentiated uterine sarcoma.^[2] Comparing with other types, ESN is much rarer and characterized as benign and inactive.^[3] Because most ESN cases rarely have any typical clinical manifestations, ESN can only be diagnosed through postoperative pathology. In this

report, a case for a 29-year-old young woman was presented, who was diagnosed with and treated for ESN in cervical parts with postsurgery complications of cervical complex adhesion atresia with extremely complicated imaging performance. During treatment, some unique suggestions were practiced for operation methods of the complex cervical adhesion and atresia. In addition, scar resection and cervical reconstruction was successfully performed. Some important experiences were also learned, which was helpful for avoiding and treating similar postoperative complications (cervical adhesions and atresia).

2. Case report

2.1. Case introduction

Consent to conduct of the case report was obtained from the Ethics Committee of Beijing Shijitan Hospital of Capital Medical University. On November 21, 2014, a 29-year-old woman was admitted as having uterine fibroid. Abdominal exploration and fibroid resection was performed under general anesthesia.

2.2. Intraoperative exploration

A nodule was found between lower part of uterus and right side of cervix to broad ligament. The nodule was about 8 cm in diameter with thick tumor nodule and a deep position. During removal of tumor pedicle, uterine segment was found penetrating into the uterine cavity. The postoperative pathological diagnosis was ESN.

After the operation, the patient suffered from amenorrhea for more than 1 year and had periodic abdominal pain for 5 times. She

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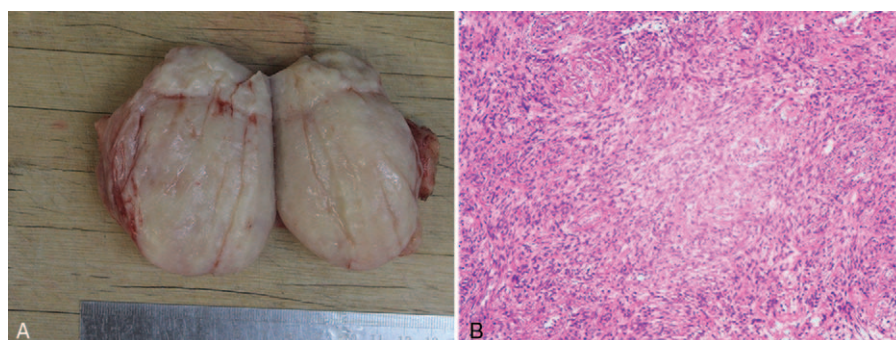


Figure 1. (A) Gross specimen of endometrial interstitial nodule: section of gray, homogeneous, delicate, border clear, and no obvious woven structure; (B) microscopic examination: spindle cell tumor, spindle-shaped tumor cells, and nuclear stained.

received ultrasound-guided cervical adhesion release on February 11, 2015 and May 22, 2015, respectively. Under the guidance of ultrasound, cervix was probed into via cervical mouth, which could not be continued at about 2 cm from cervical orifice, because front tissue was solid with great resistance. After repeated attempts, it still failed to enter the uterine cavity. During this period, oral mifepristone was given to suspend menstrual complex tide.

The patient was re-admitted because of secondary amenorrhea with periodic abdominal pain for 1 year. Magnetic resonance imaging (MRI) examination indicated separation of uterine cavity line (considering accumulation of blood) to the left after the top shift. Cervical and intrauterine condition remained abnormal. No abnormal signal was observed in cervical canal. B-mode ultrasound showed that cervical canal was not continuous in uterine cavity.

Before surgery, the patient was diagnosed with secondary amenorrhea, cervical atresia, and transcervical resection of endometrium. On December 21, 2015, the patient underwent hysteroscopy combined with laparoscopy under general anesthesia. To understand the situation of laparoscopic abdominal adhesion, B-mode ultrasound-guided hysteroscopy was performed. Laparoscopy scar resection and cervical canal reconstruction was also conducted. Preoperative cystoscopy was also performed to implant bilateral ureteral D-J tube.

2.3. Laparoscopic exploration

Normal cervical canal morphology was presented with a scar-like tissue close to the top. Ultrasound detection showed that endometrium and cervical canal line was at a right angle. Laparoscopic incision from posterior wall into uterine cavity was cut. Under guidance of hysteroscopy light, uterine fluid bled out after uterine segment scar tissue was cut.

Then, a No. 6 expanding uterus stick was put from cervix into uterine cavity, and a No. 3 neonatal tracheal catheter was placed into uterine cavity, guided by a probe. Posterior wall of the uterus incision was sutured continuously with absorbable thread. Then, the probe was removed from cervix, and catheter balloon was fixed in uterine cavity 2 cm from external orifice of cervix with injection of 1 mL saline solution. The catheter was located in uterine cavity determined by ultrasound again. The D-J tube was removed under cystoscope. The operation was smooth and menstruation was regularly restored.

Regular replacement of uterine cavity catheter was done to ensure smooth menstruation. On July 28, 2016, hysteroscopy indicated that connection of cervical canal and lower part of uterus in posterior part was not smooth. The surface was covered

with intima. The postoperative recovery was good. The indwelling catheter was removed half a year later (Figs. 1–4)

3. Discussion

Endometrial stromal nodule can occur from 23 to 86 years, and the average age is 53 years.^[2,4] No typical clinical symptoms are defined in ESN, but it often manifests as vaginal bleeding, and abdominal discomfort or pain. In addition, no specific medical imaging is defined for ESN in B-ultrasound or MRI examination.^[5] Even with more advanced methods such as diffusion-weighted magnetic resonance imaging or 18-fluorodeoxyglucose positron emission tomography, ESN presents no validated differences.^[6] Therefore, preoperative diagnosis of ESN is very difficult. ESN is often misdiagnosed as endometrial interstitial ectopic, stromal hyperplasia, or uterine fibroids.^[7] Also, it usually needs to be confirmed by postoperative pathology. In this case, pelvic mass was observed during physical examination. Then, B-mode ultrasound



Figure 2. Magnetic resonance imaging examination after removal of endometrial stromal nodule. The uterine cavity was occupied by short T1 and T2 signals, which indicated separation of the uterine cavity line (considering the accumulation of blood) with complete band. There was no abnormal signal in the myometrium with integrated uterine serosa surface. The cervix of the uterus was shifted to the upper left, and the cervical canal and the uterine cavity were continuity abnormal with abnormal cervical twist.

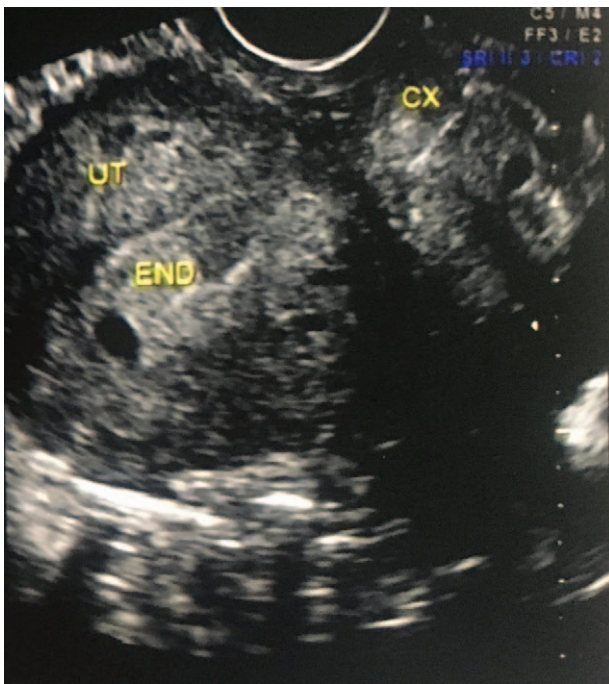


Figure 3. Endometrial interstitial nodules after removal of ultrasound prompted cervix in the uterine cavity between the continuous, intrauterine fluid dark areas.

examination indicated that a first-class echo group appeared in front of uterus, which might be cervical fibroids. In this case, preoperative diagnosis was cervical fibroids, while postoperative pathology confirmed ESN.

Postoperative pathology is essential for diagnosis of ESN. ESN is usually soft-texture, yellow or brown nodule. The general

diameter for ESN is about 7 cm, but some are up to 22 cm.^[4,8] Under the microscope, the boundary of ESN is clear.^[9] Tumor cell is similar to proliferative endometrial stromal cell with a round or spindle shape,^[9] and dense arrangement.^[11] ESN often behaves as inflating expansion growth biologically. Some tumor borders may show small irregular and finger-like performance. General protrusion is often less than 3 mm, and number of prominent is smaller than 3 without any blood vessels or lymphoid invasion.^[4,8] Immunohistochemistry is helpful in identifying endometrial stromal tumor. Single marker specificity is not strong enough for diagnosis. The combination of multiple markers can increase accuracy of diagnosis.^[10,11] The general postoperative pathology for this patient is homogeneous, delicate, and border-clear. Microscopic examination presented spindle cell tumor tissue and nuclear division 1/50 High power field, with no clear necrosis, but clear boundary. Immunohistochemistry results were listed here: A3: CD10 (part +), Ki-67 (2% +), SMA (+) caldesmon (+); A1: CD10 (+). In summary, these results are consistent with pathological manifestation of ESN.

With development of molecular genetic diagnosis technology, endometrial stromal tumor was classified as hereditary heterogeneous tumor. There is recurrence of pigment translocation, resulting in specific gene rearrangement. Among endometrial stromal tumor, the most common genetic abnormality is caused by (7; 17) (p15; q21) translocation fusion of the JAZF1-SUZ12 gene.^[12-14] Ability of distinguishing endometrial stromal tumor was improved by involving genetic alteration.

The patient was a 29-year-old young woman. The auxiliary methods (ultrasound, radiation, etc) was adopted for post-operation follow-ups. During the postoperative clinical follow-ups, the patient experienced no menstruation, but reported cyclical abdominal pain 5 times. Imaging examination showed cervix and uterine cavity continuity disorder with abnormal cervical distortion, considering occurrence of complex cervical

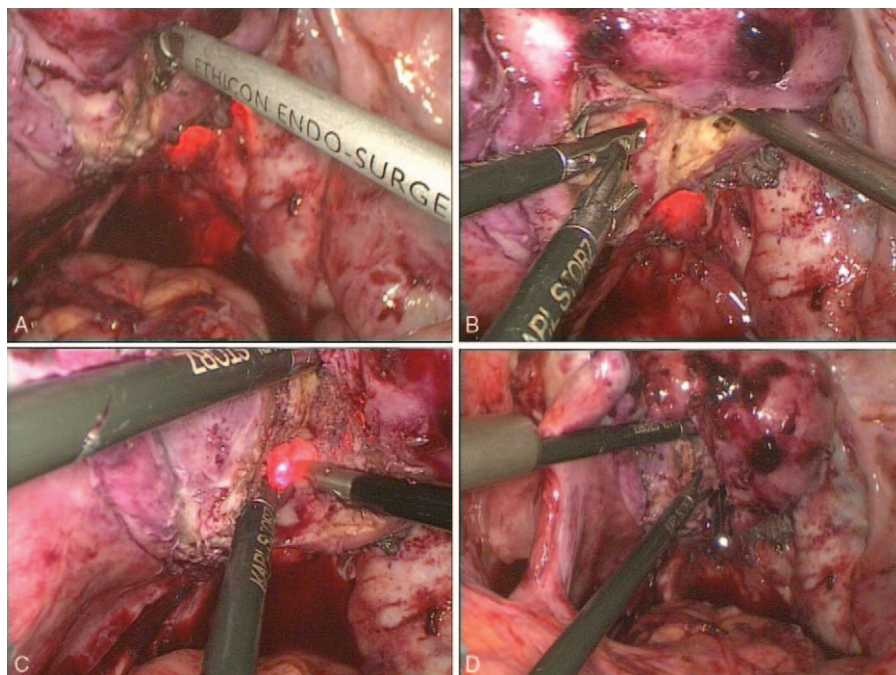


Figure 4. Hysteroscopy combined with laparoscopy process (scar resection and cervical reconstruction). (A) Cervical scar like tip is closed; (B, C) laparoscopic uterine incision from posterior wall into the uterine cavity, the lower uterine segment scar tissue was cut under the guidance of hysteroscopic light; (D) the 6 expanding uterus stick was replaced into the uterine cavity.

adhesion. The endometrial stromal lesion for this patient was located in lower part of uterus and cervical junction close to endometrium. Because endometrial internode nodules are special, postoperative complications occurred easily. During stripping process, postoperative wound bleeding and inflammation happened commonly. Surgery can also lead to local anatomical structural disorder, scar traction,^[15] and other abnormal cervical malformation.

In this case of cervical adhesion, the imaging performance is extremely complex angular deformity. Through nuclear magnetic imaging, cervix and palace presented as a “Twisted and Angled Staircase” image, which was very rare. To solve the clinical symptoms of the patient, specificity of the complications was also taken into consideration. A careful preoperative multidisciplinary discussion was conducted, and 6 choices of surgical programs were as follows: (1) direct stitching with vaginal after resection of cervix; (2) attempt to get up and down to reset cervix; (3) destructive treatment, opening violently by 45 degrees, from top to bottom; (4) open surgery from top to bottom, starting at bottom of uterus; (5) open surgery at abdomen; and (6) investigation of adhesion by laparoscope, and hysteroscopy was operated with assistance of B-mode ultrasound, which can be explored via intra-cavity ultrasound, while ultrasound monitored up and down during conduction. After feasibility of the above 6 programs and fact that the patient’s cervix has deformed with multiple adhesion scars was discussed, which meant that the patient has lost normal anatomical structure, the last surgical plan was chosen to decrease difficulty in restoring original anatomy and to avoid recurrence of other organs to reduce surgical trauma.

Studies^[16,17] reported that silicone drainage tube and Foley catheter puncture needle through center of uterus and cervix could be used, leaving the end of the drainage tube attaching to vagina and vestibule, to solve the problem of menstruation. Use of uterine type intrauterine device under 18 silicone catheter could play a fixed anti-off role and support formation of cervical cannula to prevent adhesion, narrowing, and atresia.^[18] For this case, a comprehensive presurgery assessment was conducted to determine the use of ultrasound-assisted subcultural uterine laparoscopic reconstruction of the tunnel, from top to bottom, and the drainage tube from vagina through cervix into uterine cavity. Ultrasound-guided exploration and expansion of cervix was also adopted to avoid blind operation and damage caused by scope. A number of literatures reported that reconstruction of tunnel can easily cause atresia re-adhesion, so catheter within uterus was essential to prevent re-adhesion. The catheter within uterus was a support organization and played a role in fluid drainage. In addition, to avoid postoperative infection, antibiotics were used and the catheter was replaced regularly. The surgery achieved desired results.

4. Conclusions

In conclusion, this patient is diagnosed with ESN, which is a rare disease. The ESN grows at a special site and causes postoperative complications easily. Firstly, the patient’s nodule position is low, the size is huge, and the intraoperative incision is long, requiring the penetration from the lower uterine segment through the uterine cavity to the cervix. The lower uterine segment and cervix should be paid attention during suturing in this situation. Strict hemostasis and anatomical morphology maintenance can help to avoid formation of stenosis, distortion, or postoperative adhesion. A catheter or silicone drainage tube can be stitched in uterine cavity before suturing, so the cervical tube leads

through the vagina from the lower uterine segment.^[19] The drainage tube can be replaced by a stent 1 or 2 weeks later to reduce postsurgical adhesion and deformity healing, and to avoid cervical distortions. Secondly, close and positive follow-ups should be planned after the endometrial stromal resection. The follow-ups can detect problems early, which contribute to avoid late-detected trauma with harder surgical interventions caused by scars and hyperplasia. Thirdly, the patient has abnormal cervical distortion with angular deformity. Before the cervical reconstruction, a full assessment of imaging is required. MRI is preferred because it has a good resolution of soft tissue and a detailed distinction between the palace, cervix, and vaginal segment, which can present the shape of the cervix and cervical canal, and also the relationship with the surrounding tissues, which is very helpful during the determination of surgical programs. Fourthly, the patient had lost normal anatomy of the cervix. It is difficult to force restoring the normal anatomy and can damage the surrounding organs easily. So, the reconstruction of the tunnel is a solution for the problem of menstruation.

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