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Magnetic Resonance Imaging of Cellular Blue Nevus of the Uterus Diffusing To the Pelvic Cavity: A Case Study

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Abstract

Blue nevi of the uterus and uterine cervix are rare. We presented here a case of a 26-year-old patient who had history of vaginal bleeding and menorrhagia. MRI of the patient showed that the middle and lower uterine segments as well as the endocervix were replaced by abnormal tissues with marked thickening. Fat-saturated series showed that there were multiple focal high-intensity signals on T1WI and low-intensity signals on T2WI in the lesion. Simultaneously, there were similar abnormal tissues distributed in the right uterine horn, the left pelvic wall, the outer serosal layer of the posterior wall of the bladder and the space between the bladder and the uterus, etc. Biopsy of uterine tumor and partial resection of pelvic tumor were performed, and it was pathologically confirmed to be cellular blue nevus.

Keywords: Blue Nevus; Magnetic Resonance Imaging; Uterus; Diffusion

Background

Blue nevus is a benign melanocytic skin tumor, which is rare in extra cutaneous sites. Blue nevus is also visible in cervix, vagina, spermatic cord, prostate, hilar, eyelid, eye, oral mucosa, esophagus, maxillary sinus and lymph nodes, etc. [1-3]. Among them, cervix is an extra cutaneous organ with the highest incidence rate of blue nevus, but it is not easy to be detected by examination due to the small lesion [4], these lesions are hardly detected by cross-sectional imaging alone. MRI of the patient suggested that there were tumors in the uterus and uterine cervix. Furthermore, there was high-intensity signals manifestation of pelvic diffusion.

We obtained informed consent from the patient for the clinical information used in this study, and this study had been approved by the Ethics Committee of our hospital.

Case Report

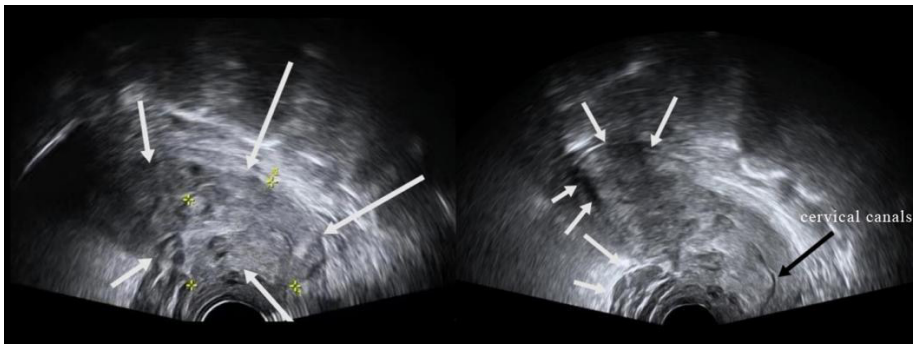

Figure 1: Transvaginal two-dimensional Doppler Ultrasound. There were continuously uneven echoes from the uterine cervix to the middle and lower uterine segments with the thickest part of 3.5cm and the cervical canals (pointed by black arrows)

This study reported a case of a 26-year-old housewife who visited our hospital due to heavy menstrual bleeding and anemia. It was known by medical history inquiry that the patient suffered from dark-purple cervical mucosa by gynecological examination in Guangze County Hospital 6 years ago, but no treatment was conducted. After that, the patient got pregnant, but black soft tissues were found in the uterine cervix when underwent cesarean section in Guangze County Hospital in August 2014 (the patient cannot

provide complete information). Laboratory test in our hospital showed that hemoglobin was 89.0g/l, and other indicators showed no abnormality. Transabdominal and transvaginal color Doppler ultrasonography found that there were continuously uneven echoes from the uterine cervix to the middle and lower uterine segments, and there were rich colorful blood flow signals with the thickest part of about 3.5cm (Figure 1). Multiple hypoechoic nodules were detected besides the uterus and in the space between the uterus and the bladder. Some of the nodules aggregated in flaky shape. Range of the hypoechoic area between the bladder and the uterus was about 4.2×1.5×5.6cm, and there was no obvious boundary with the bladder.

There were continuously uneven echoes from the uterine cervix to the middle and lower uterine segments with the thickest part of 3.5cm

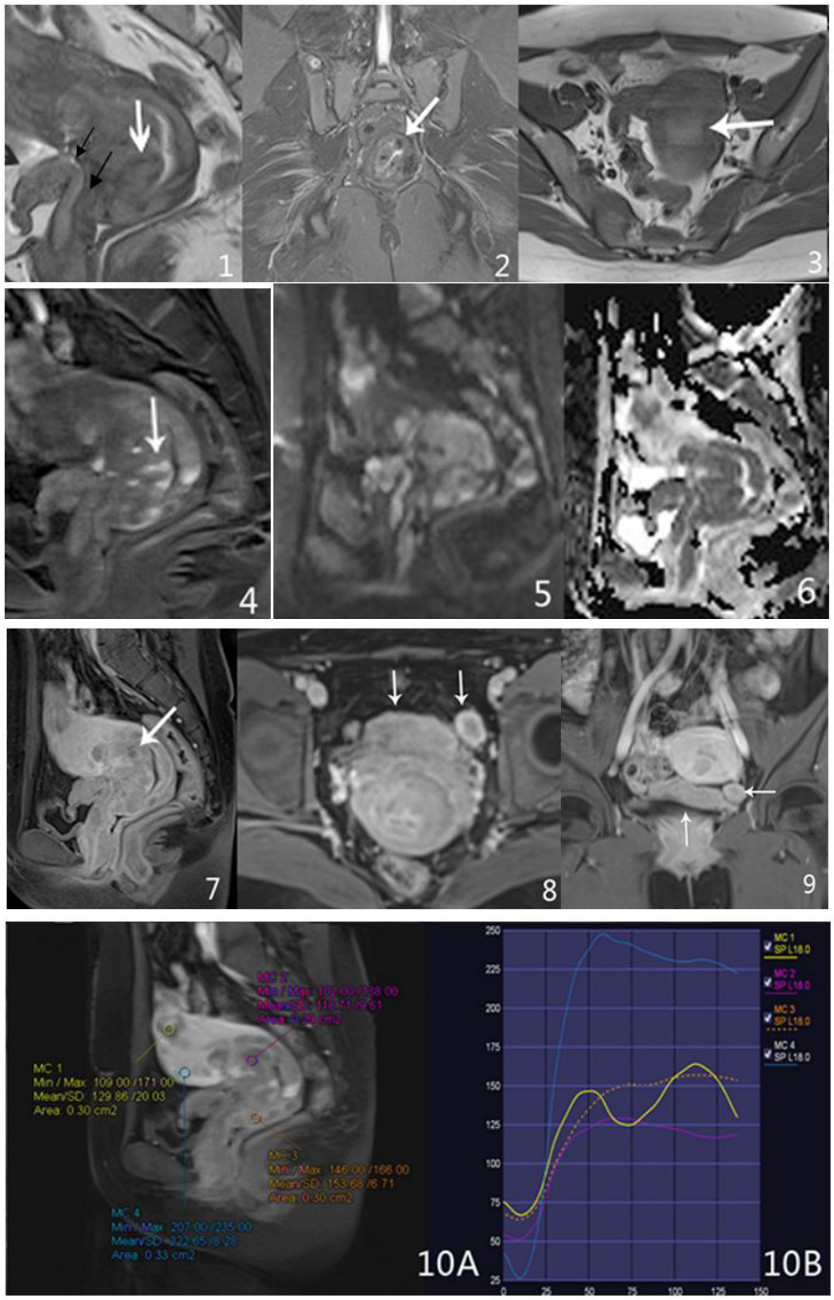

Figure 2: MRI examinations performed with 3.0T superconducting MRI systems

1) Sagittal T2-WI showed that there was abnormal soft-tissue shadows distributed from the uterine cervix to the lower uterine segment, as well as mixed signals slightly higher than those of myometrium. There were multiple flaky low-signal nests (pointed by white arrows). Moreover, structure of the cesarean section area in the lower uterine segment was unclear, tumor tissues were rich, and the junctional zone was incomplete. Meanwhile, soft-tissue shadows of the same signals can be observed in the right uterine horn, outer serosal layer of the bladder as well as the space between the bladder and the uterus (pointed by black arrows); **2)** Coronal image on FS-T2W1 clearly showed that there were extremely low-signal nests in the lesion (pointed by arrows); **3)** Transverse image on T1WI showed that the lesion (pointed by arrows) was higher than signals of myometrium; **4)** Lesion had signals that were isointensity or slightly higher compared to lesions showed on FS-T1W1 in the same positions in Figure 2-1. The scattered flaky high-signal nests corresponded to low-signal nests in Figure 1 (pointed by arrows); **5,6)** Sagittal DWI (b = 800 s/mm^2) and ADC images corresponded to those in the same positions in Figure 2-1, indicating that lesion diffusion was restricted, which manifested as increased signals and

decreased ADC value; **7)** Enhanced scan from the sagittal position showed that the lesion was unevenly enhanced, and signal intensity was lower compared to myometrium; **8,9)** Enhanced transversal and coronal images showed that enhancement of pelvic nodules and subserosal lesion of the balder were similar to cervical lesion (pointed by arrows); **10)** Dynamically enhanced images; Lesion in **A** (positioning image) showed on **B** (time-signal intensity curve) was plateau-type signals (red, yellow, orange), and the intensity was lower compared to myometrium (blue)

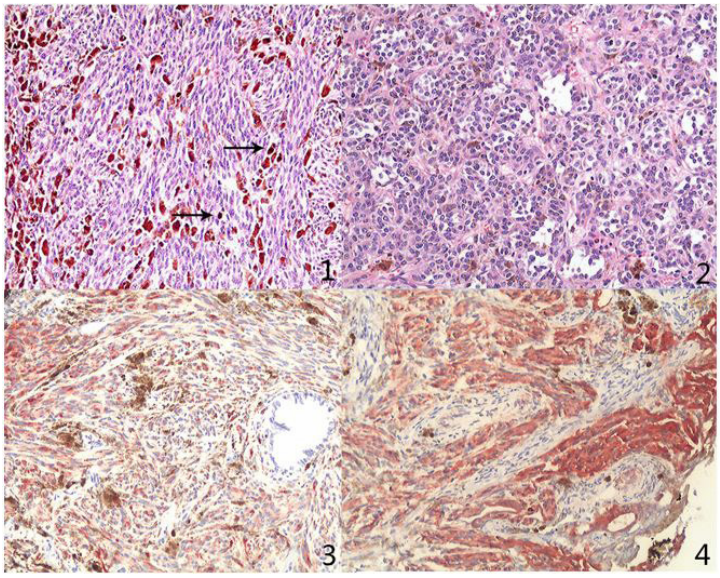

Figure 3: The isolated specimens sent to examination

1) HE staining (x200) showed that spindle cells containing black pigment were arranged in bundle, and the nuclei were regular (pointed by arrows); **2)** HE staining (x200) showed that the epithelioid tumor cells were nested, and there were melanin particles in the cytoplasm; **3,4)** HMB45 and MelanA with strongly positive expression

MRI examinations were performed with 3.0T superconducting MRI systems (Skyra, Siemens Healthcare). Images showed that there were abnormal signal shadows with various thicknesses extensively distributed from the endometrium to the endocervix, and the thickness ranged from 0.9cm to 3.4cm. Junctional zones of some parts were incomplete, and structure of the scar site in the lower uterine segment was unclear (Figure 2-1). The lesion had mixed and slightly high-intensity signals on T2W1, and focal (nested) low-intensity signals were scattered (Figure 2-1). Low-intensity signals in the lesion were more clearly displayed on fat-suppressed T2WI (Figure 2-2). Signals on T1WI were generally higher compared to the myometrium (Figure 2-3). There were multiple high-signal nests in the lesion on lipid-suppressed T1WI (Figure 2-4), which corresponded to low-signal nests on T2WI. The lesion showed high-intensity signals on DWI (b value of 800s/mm^2) (Figure 2-5). ADC value was measured to be 0.644×10^{-3}mm^2/s, which were in low-intensity signals (Figure 2-6). After administration of contrast medium (Gadopentetate dimeglumine injection, Beijing BeiLu Pharmaceutical Co Ltd), enhanced scan showed that there were unevenly enhanced signals in the lesion, and the intensity was lower compared to the myometrium (Figure 2-7). Masses with the same manifestations were distributed in the right uterine horn, small nodules of the left pelvic wall, the posterior wall of the bladder and the space between the bladder and the uterine (Figure 2-1, 2-8 and 2-9). Dynamically enhanced time-signal intensity curve of the tumors was in plateau shape, and the intensity was lower compared to the myometrium (Figure 2-10).

Subsequent gynecological examination showed that the uterine cervix was violet-blue in color, the endocervix was uneven under a hysteroscope, and the violet-blue masses adhered to the uterus and endocervix as oil paint, leading to stiff uterine wall. Thus, biopsy of tumor tissue was performed, and the pathological results showed that it was cellular blue nevus. Ten days later, the gynecologist decided to keep the uterus after communicating with the patient, and pelvic tumor resection was performed by cooperating with an urologist. During the operation, a blue columnar mass with the size of about 2×3cm was observed at the right uterine horn, which was not resected. Dark-purple masses with various sizes were observed in the peritonel reflection of the bladder, lower uterus, posterior wall of the bladder and the lateral pelvic wall, and most of which were aggregated as a whole part with the range of about 8×9cm. The surface of the masses was uneven and smooth without adhesion, and the texture of the masses was tough. The masses were separated and resected, and a black nodule with the size of about 2×3cm on the left pelvic wall was also resected to be sent for pathological examination. Pelvic examination showed that multiple brown or black dotted lesions were scattered in Douglascul-de-sac, bilateral sacrospinous ligament and serosal surface of rectum, which were not resected.

The isolated specimens sent to examination were grayish black/taupe brown and medium in texture, with thin coating in some areas and a small amount of fiber segregation inside. Tumor cells arranged in either bundle, nest and acinar under a light microscope, and fibro vascular separation was visible locally. The tumor cells were either fusiform, fat fusiform or epithelial, transitional areas were detected, and cytoplasm was eosinophilic or transparent. There were melanin particles in the cells. Moreover, the cells were not obviously atypia, the nuclei were either oval, round or slender, the nuclear membrane was regular, the chromatin was evenly

distributed, the nucleolus was not obvious or small, no pathological mitotic figure was observed, and there was no hemorrhage and necrosis in tissues (Figure 3-1 and 3-2). Immunohistochemical examination showed that both HMB45 and MelanA were strongly positive (Figure 3-3 and 3-4), SOX-10 was positive locally, while S-100 and CyclinD1 were negative. Moreover, Ki-67 index was 1%. Molecular pathology indicated that there were no V600E mutant loci in BRAF gene. The final pathological report showed that the tumor was cellular blue nevus. Moreover, proliferation activity of tumor cells was low, and the morphology was mild, but growth of tumor cells was invasive, thus we recommend the patient to have clinical follow-up.

Discussion

The blue nevi are pathologically divided into two types: common blue nevus and cellular blue nevus [3-5]. Cellular blue nevus manifests as fusiform melanocytes arranged in bundle or nest, which aggregate to form nodules with clear boundary. Common blue nevus manifests as melanin-deficient fusiform melanocytes and melanin-rich dendritic melanocytes, as well as collagen bundles between cells, which aggregate to form nodules with unclear boundary [3]. In our case, typical melanin-rich fusiform melanocytes were observed under a microscope. The melanocytes were not atypia, and there was no genetic mutation. Results of immunohistochemistry demonstrated that it was benign cellular blue nevus. Uterine cervix is the most predilection extra cutaneous site of blue nevus [4,6]. The first case of blue nevus of the uterine cervix was reported in 1922 [2,7]. However, blue nevus in the endometrium is extremely rare, which can be found accidentally due to endometrial polyps or tumors [4]. In our study, the young patient had clinical manifestation of anemia due to heavy menstrual bleeding, which may be correlated with the extensive involvement of the endometrium by tumors. At the same time, cases with blue nevus in the endometrium and uterine cervix are extremely rare [4]. Therefore, this case attracted the author's attention.

MRI is the best pelvic examination method for women, who cannot only display the anatomical relationship of lesions in multiple views, and the application with multiple parameters can provide histological characteristics of lesions. Therefore, MRI is of active significance in disease diagnosis. In this study, tumor tissues with various thicknesses were diffusely distributed from the middle and lower uterine segments to the endocervix. Furthermore, there was no liquefaction necrosis, and the junctional zone was basically complete, indicating that tumor growth was mild (benign characteristic). Structure of the lower uterine segment was unclear with rich tumor tissues, which was considered to be caused by surgery. The rich pigmented component of tumor resulted in signal characteristics that were different from common tumors, manifesting as multiple small plaques with paramagnetic short T1 and short T2 signals. The sign can be considered to be characteristic MRI performance of tumor rich in melanin [8]. Enhanced scan of the lesion showed that the lesion was moderately evenly enhanced, and the time-signal intensity curve was in a plateau shape, suggesting that blood supply of the tumor tissues was abundance, because the tumor was hypo enhancing on post contrast T1WI compared to the myometrium and this was confirmed by time-intensity curve to have lower peak compared to the myometrium.

Imaging report of blue nevus of the uterine cervix is rare, which may be because the lesion is usually less than 5mm, and locate in submucosal stroma but not protrude from the mucosal surface, thus it is not easy to be detected [2,4,9]. Blue nevi develop both in the endometrium and uterine cervix is rare [4]. In this case, the lesion and the range were large, and there were no similar cases reported previously. Author of this study speculated that tumor diffusion may be directly correlated with cesarean section, in which tumor tissues were exposed at the incision due to surgery, leading to the tumor infiltrate to vesicouterine pouch and outer serosal layer of the bladder through the incision at the lower uterine segment. Multiple nodules in the pelvic cavity were suspected to be the growth of tumor cells shed during the cesarean section, which formed multifocal lesions, thus it was easily to be misdiagnosed as a malignant tumor before the operation.

In this case, characteristic manifestations of multiple paramagnetic nested nodules on MRI were easily to be differentiated from tumors such as cervical cancer, endometrial cancer, leiomyosarcoma and lymphoma, etc. [8], but it was difficult to be differentiated from malignant melanoma [4,8,10]. Therefore, pathological examination was needed for final diagnosis.

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