MRI FINDINGS OF COTYLEDONOID DISSECTING LEIOMYOMA OF THE UTERUS

Shigenobu Motoshima,[1] Hiroyuki Irie,[2] Takahiko Nakazono,[3] Fumio Yamasaki,[4] Yoshifumi Nakao[5]

[1] Department of Radiology, Takagi Hospital, Japan.
[2] Department of Radiology, Faculty of Medicine, Saga University, Japan.
[3] Department of Radiology, Saga Social Insurance Hospital, Japan.
[4] Department of Pathology, Faculty of Medicine, Saga University, Japan.
[5] Department of Obstetrics and Gynecology, Faculty of Medicine, Saga University, Japan.

PJR July - September 2011; 21(3): 134-138

ABSTRACT

Cotyledonoid dissecting leiomyoma of the uterus is an extremely rare smooth muscle tumor and is classified as leiomyoma with growth pattern variants. We report a case of cotyledonoid dissecting leiomyoma of the uterus, and discuss the diagnostic MRI findings, including diffusion-weighted MRI and contrast-enhanced dynamic MRI.

Key words: Magnetic Resonance Imaging; Diffusion Magnetic Resonance Imaging; Dynamic MRI; Gadolinium DTPA; Uterus; Fibroid

Introduction

Cotyledonoid dissecting leiomyoma of the uterus is an extremely rare, benign smooth muscle tumor of the uterus, classified as a leiomyoma with growth pattern variants in the World Health Organization (WHO) histological classification of the tumors of the uterine corpus.[1,2] Roth et al.[1] designated four cases of these tumors as "cotyledonoid dissecting leiomyoma" of the uterus in 1996. Conventional uterine leiomyomas are symmetrical, expansile, rounded, and circumscribed, while Roth et al.[1] focused on the appearance of the exophytic component, which is deep red and bulky, resembles cotyledons of the placental tissue and extends from the lateral uterine surface in the region of the cornu. These tumors are connected with the endophytic component of the dissecting patterns in the uterine myometrium by fascicles of neoplastic smooth muscle.

Case Report

A 39-year-old nulliparous woman presented feeling an abdominal mass for 6 months, and an intrapelvic mass was found at a local hospital. The mass was suspected to be an ovarian malignant tumor, and she was referred to our institution. Pelvic MRI showed a solid mass on the dorsal side of the uterine corpus, which filled the whole pelvic cavity, maintaining the existing structures. On a T2-weighted image (T2-WI), both ovaries were identified as separate from the mass, and there were flow voids between the mass and uterine corpus (Fig. 1 A, B).

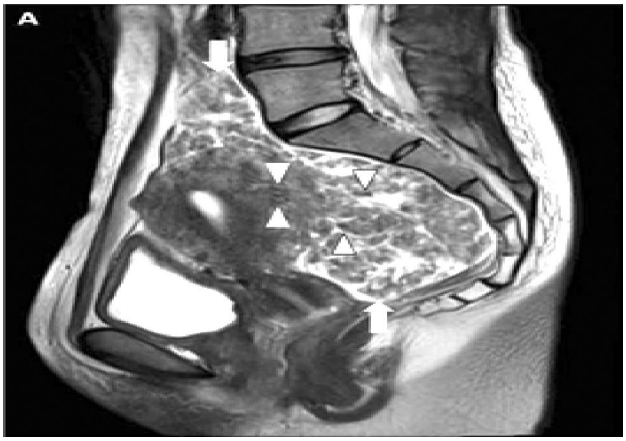

Figure 1-A: Sagittal T2-weighted fast spin-echo MRI (T2-WI) shows a solid mass (arrows) on the dorsal side of the uterine corpus. There are flow voids (arrowheads) between the mass and uterine corpus. The solid mass shows heterogeneous high signal intensity, and iso-signal intensity multinodular structures are seen in the mass. The perinodal area in the mass shows reticular high signal intensity.

Correspondence : Shigenobu Motoshima
Department of Radiology, Takagi Hospital
141-11, Sakemi, Okawa, Fukuoka 8310016,
Japan. Tel: +81-944-87-0001
E-mail: s-motoshima@gmail.com

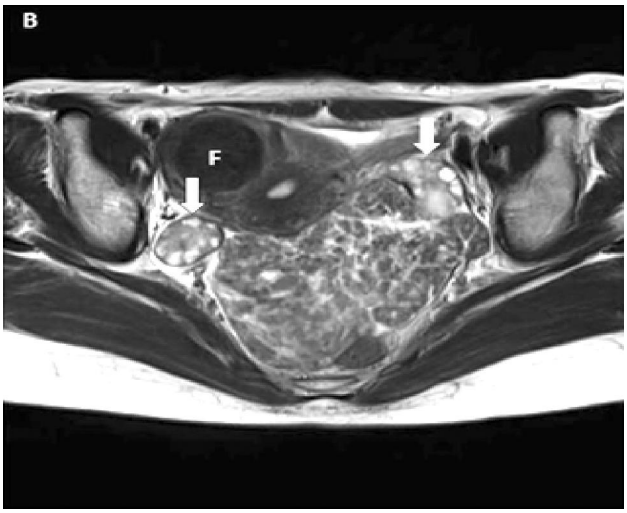

Figure 1-B: On axial T2-WI, both ovaries are identified as separate from the mass (arrows). Ordinary uterine fibroid (F) is seen in the anterior wall of the uterine myometrium.

The mass was suspected to be continuous with the dorsal side of the uterine cornu. The solid mass showed heterogeneous high signal intensity compared to the outer myometrium, and iso-signal intensity multinodular structures were seen in the mass. The perinodal area in the mass showed reticular high signal intensity on T2-WI. Hemorrhage or necrosis was not identified in the mass. On DWI (b= 1000 s/mm^2), the mass showed iso-signal intensity compared to the outer myometrium, and the apparent diffusion coefficient (ADC) map did not show restricted diffusion in the mass. The ADC values of the mass were 1.9x10^{-3} mm^2/s (Fig. 2 A,B).

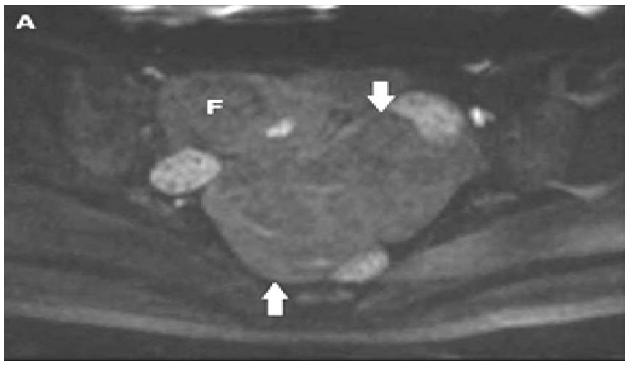

Figure 2-A: On axial diffusion-weighted echo-planar MRI (DWI) (b =1000 s/mm^2), each nodule of the mass (arrows) shows iso-signal intensity.

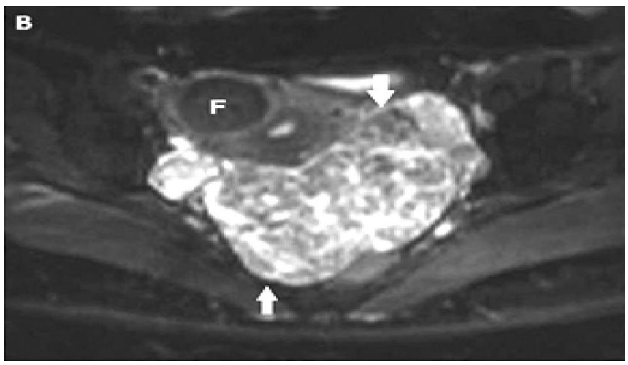

Figure 2-B: On an apparent diffusion coefficient (ADC) map, the ADC values of the mass is 1.9x10^{-3} mm^2/s. F= fibroid.

On contrast-enhanced dynamic MRI, each nodule showed extremely weak, gradually increasing enhancement compared to the outer myometrium (Fig.3 A, E).

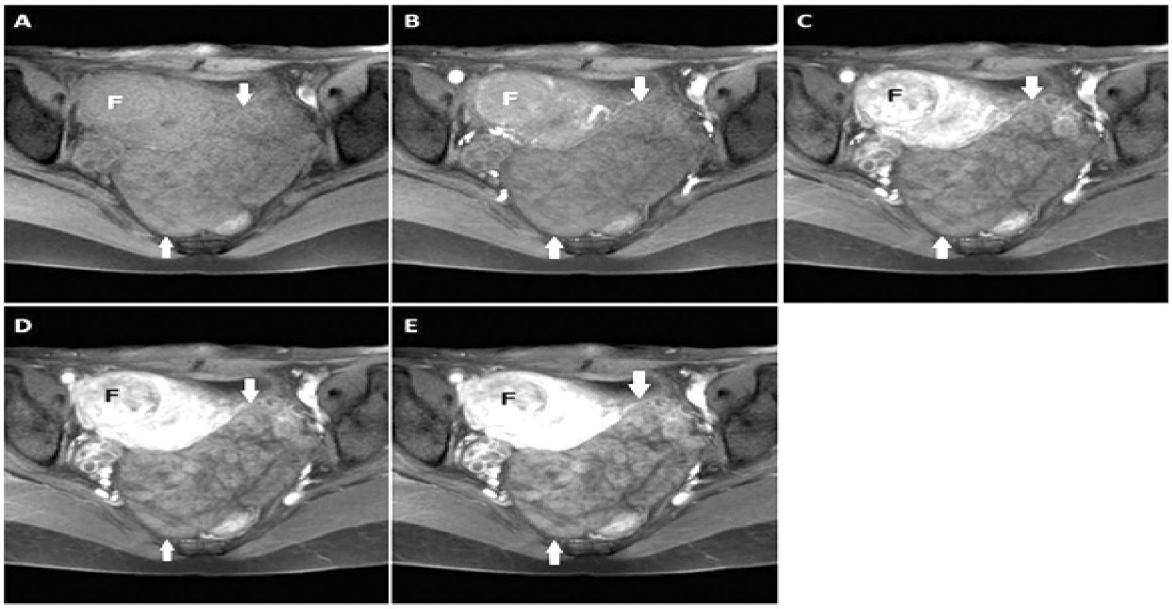

Figure 3A-E: On contrast-enhanced dynamic MRI (A 0 seconds B 30 seconds C 60 seconds D 90 seconds E 120 seconds), each nodule in the mass (arrows) shows extremely weak, gradually increasing enhancement. F= fibroid.

No retroperitoneal lymph node swelling or peritoneal dissemination was found. Small amounts of ascites were seen in the pouch of Douglas. There were no likely suggestions of a malignant tumor, and we speculated that the tumor arose from the uterine mesenchyme. We preoperatively diagnosed a benign uterine smooth muscle tumor, which showed an atypical morphological development. Surgical exploration confirmed a pinkish, solid polypoid mass, which arose from the posterior surface of the uterine corpus into the pelvic cavity (Fig. 4A). The polypoid mass was attached to the posterior aspect of the uterine corpus. The morphology of the mass was discoid, formed of numerous finger-like or bulbous protrusions (Fig. 4B). Both ovaries and fallopian tubes appeared unremarkable. The pathological diagnosis of a frozen section was leiomyoma. Conservative surgery was performed for the purpose of preserving fertility; the tumor was resected including the pedicle and an approximately 1.5-cm depth of the uterine myometrium. The polypoid mass was sized 14.0 x 14.0 x 6.0cm and pathological examination revealed multinodular proliferating smooth muscle cells which did not show a significant cytological atypia or coagulative necrosis (Fig. 4C).

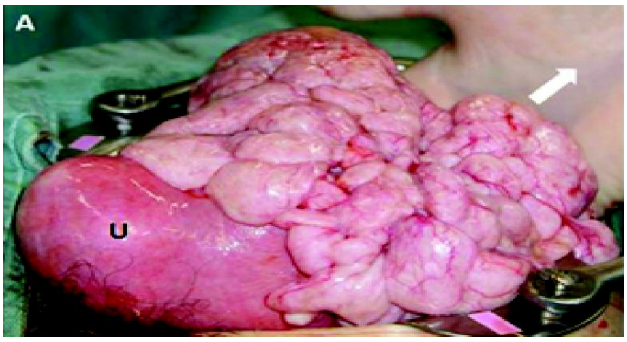

Figure 4-A: A pinkish, solid polypoid mass arises from the posterior surface of the uterine corpus into the pelvic cavity. Arrow shows the cephalic direction. U= uterus.

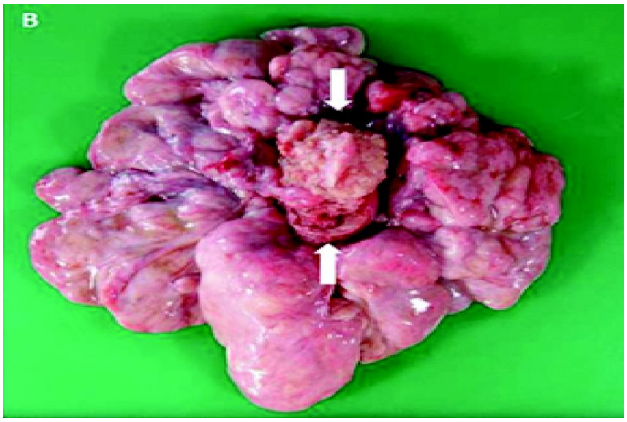

Figure 4-B: Resected specimen including pedicle (arrows) shows a discoid mass formed of numerous finger-like or bulbous protrusions.

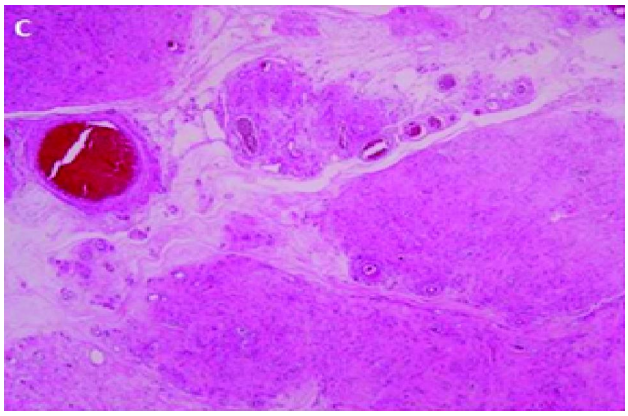

Figure 4-C: Pathological examination (Hematoxylin and eosin stain ×20) reveals multinodular proliferating smooth muscle cells which do not show a significant cytological atypia or coagulative necrosis. Regions of edema and hyaline degeneration are seen in the nodules, and the perinodal stroma shows a highly hydropic degeneration containing congested vessels.

Regions of edema and hyaline degeneration were seen in the nodules, and the perinodal stroma showed a highly hydropic degeneration, containing congested vessels. Intravenous development of a tumor was not shown microscopically. The gross appearance of the tumor resembled the maternal surface of placental tissue and was diagnosed as a cotyledonoid dissecting leiomyoma of the uterus. The patient was free from findings of tumor regrowth at a 36-month postoperative follow-up.

Discussion

Approximately 20 cases of cotyledonoid dissecting leiomyoma have been reported in the literature. These cases ranged in age from 23 to 41 year-old, and the tumors ranged in size from 10 to 25 cm.[3,4] To explain the deep red color of the exophytic components, it is hypothesized that the tumor impedes venous drainage as it grows in cotyledonoid patterns beyond the confines of the uterus and congestion develops.[1] One possible explanation for this characteristic extension of the tumor is that the tumor extends along muscular vessels from the stratum vasculare of the uterine corpus into the connective tissue of the broad ligament, since there is no anatomical barrier.[1] The endophytic component shows tongues of abnormal smooth muscles that extend significantly from the parent leiomyoma between the fascicles of the adjacent myometrium, which was characterized by ill-defined borders.[5]

Accurate preoperative assessment of uterine lesions and the characterization of them as benign or malignant can be very important to narrow the differential diagnoses in order to avoid overtreatment, especially

in fertile women for whom conservative surgery might be considered.[6] Morphologically, cotyledonoid dissecting leiomyoma frequently arises from the subserosal myometrium of the lateral border of the uterus and extends into the broad ligament and pelvic cavity, and the intramural component invades the fascicles of the myometrium. Owing to its bizarre, sarcoma-like gross appearance, cotyledonoid dissecting leiomyoma has been diagnosed as a malignant tumor, although the tumors of the reported cases have shown no malignant behavior or recurrence on follow-up.[4] Differential diagnoses of the heterogeneous ill-defined endophytic component in continuity with the exophytic component of the uterus may include uterine leiomyosarcoma, low-grade endometrial stromal sarcoma, and intravenous leiomyomatosis. Preda et al.[6] reported MRI findings of a case of cotyledonoid dissecting leiomyoma, and the tumor showed heterogeneous intensity with patchy iso-signal intensity nodules and thin high signal intensity stromal components on T2-WI compared to muscle. These MRI findings are similar to those in our case. In our case, reticular high signal intensity in the mass on T2-WI reflected hydropic degeneration of the perinodular stroma on pathological findings. This MRI finding may be the characteristic of cotyledonoid dissecting leiomyoma.

Leiomyosarcoma shows high signal intensity on T2-WI, while small areas of high signal intensity are seen within the tumor on T1-WI, and there are some unenhanced pocket-like areas after the administration of contrast materials which reflect intratumoral hemorrhage and coagulative necrosis. One morphologic feature of low-grade endometrial stromal sarcoma is that the myometrial lesion shows continuous extension into the adjacent structures along the fallopian tubes and surrounding uterine ligaments. This finding reflects vascular and lymphatic involvement of the tumor.[7] A characteristic imaging feature of low-grade endometrial stromal sarcoma is the presence of bands of low signal intensity on T2-WI within the area of myometrial invasion.[7]

Tamai et al.[8] reported that DWI and ADC values measurement may have the potential to differentiate uterine sarcomas such as leimyosarcomas and endometrial stromal sarcomas from benign leiomyomas. In general, malignant tumors have higher cell density than benign tumors and show high signal intensity on DWI and low ADC values on ADC map. In our case, the tumor was initially suspected to be a malignant tumor but did not show restricted diffusion on DWI. This MRI finding was useful for the pre-operative diagnosis and may reflect low cell density and edematous stroma in the tumor that were observed the pathological findings.

The usefulness of contrast-enhanced dynamic MRI in the differential diagnosis of leiomyosarcoma from ordinary leiomyoma has been reported.[9] Goto et al.[9] reported that degenerated leiomyoma shows slight or irregular enhancement on contrast-enhanced dynamic MRI. Leiomyosarcoma shows rapid enhan-cement in the early phase (at 20-90 seconds), and the myometrium is enhanced gradually in the later phase (at 120-180 seconds) on contrast-enhanced dynamic MRI.[9] In our case, the contrast-enhanced dynamic MRI finding was extremely weak, with gradually increasing enhancement compared to the outer myometrium. This MRI finding may reflect the pathological findings that the blood flow of the tumor was decreased due to congestion.

Intravenous leiomyomatosis has multiple myometrial masses or a lobulated myometrial mass, which is a wormlike plug tumor that extends within the veins of the broad ligament.[10] The infiltration pattern of cotyledonoid dissecting leiomyoma may be similar to that of intravenous leiomyomatosis.[2] Intravenous leiomyomatosis may also show marked hydropic degeneration.[10] A rigid distinction between intravenous leiomyomatosis and cotyledonoid dissecting leiomyoma may be difficult to make based on the morphological and lesion characterization on MRI.

Clinicians and radiologists should be alert to the presence of this rare disease entity and should recognize the characteristics of imaging findings and differential diagnoses. In our case, MRI findings reflected the pathological findings of this rare tumor. In addition to the morphological imaging assessment, comprehensive evaluation of each sequence including DWI and contrast-enhanced dynamic MRI may be useful to differentiate cotyledonoid dissecting leiomyoma from other malignant tumors, especially uterine sarcomas. Being familiar with the MR imaging diagnosis of this disorder may help to determine the precise management strategy, including fertility preservation surgery.

Acknowledgments: The authors acknowledge the invaluable assistance of Yuka Okajima, M.D., Mari Tanaka, M.D., and Yosuke Hagio, M.D., Ph.D.

References

1. Roth LM, Reed RJ, Sternberg WH. Cotyledonoid dissecting leiomyoma of the uterus: the Sternberg tumor. Am J Surg Pathol 1996;**20:**1455-61.

2. Hendrickson MR, Tavassoli FA, Kempson RL, McCluggage WG, Haller U, Kubic-Huch RA. Mesenchymal tumours and related lesions. In Tavassoéli FA, Devilee P, eds: Pathology and genetics tumours of the breast and female genital organs, World Health Organization classification of tumours. Lyon: IARCP Press, 2003;233-44.

3. Mi-Jin Kim, Yoon-Ki Park, Jae-Ho Cho. Cotyledonoid dissecting leiomyoma of the uterus: a case report and review of the literature. J Korean Med Sci 2002;**17:** 840-4.

4. Driss M, Zhioua F, Doghri R, Mrad K, Dhouib R, Romdhane KB. Cotyledonoid dissecting leiomyoma of the uterus associated with endosalpingiosis. Arch Gynecol Obstet 2009;**280:**1063-5.

5. Roth LM, Reed RJ. Dissecting leiomyomas of the uterus other than cotyledonoid dissecting leiomyoma: a report of eight cases. Am J Surg Pathol 1999;**23:**1032-9.

6. Preda L, Rizzo S, Gorone MS, Fasani R, Maggioni A, Bellomi M. MRI features of cotyledonoid dissecting leiomyoma of the uterus. Tumori 2009; **95:** 532-4.

7. Koyoma T, Togashi K, Konishi I, Kobayashi H, Ueda M, Kataoka ML, et al. MR imaging of endometrial stromal sarcoma: correlation with pathologic findings. Am J Roentgenol 1999; **173:** 767-72.

8. Tamai K, Koyama T, Saga T, Morisawa N, Fujimoto K, Mikami Y, et al. The utility of diffusion-weighted MR imaging for differentiating uterine sarcomas from benign leiomyomas. Eur Radiol 2008;**18:** 723-30.

9. Goto A, Takeuchi S, Sugimura K, Maruo T. Usefulness of Gd-DTPA contrast-enhanced dynamic MRI and serum determination of LDH and its isozymes in the differential diagnosis of leiomyosarcoma from degenerated leiomyoma of the uterus. 2002;**12:** 354-61.

10. Clement PB, Young RH, Scully RE. Intravenous leiomyomatosis of the uterus: a clinicopathological analysis of 16 cases with unusual histologic features. Am J Surg Pathol 1988;**12:** 932-45.