Case Report

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Cotyledonoid dissecting leiomyoma: an uncommon form of a common disease

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Leiomyomas are benign uterine smooth muscle neoplasms with varied morphology that are well known to undergo secondary changes. Cotyledonoid dissecting leiomyoma is a rare and distinct form of leiomyoma that poses a diagnostic challenge for clinicians, radiologists, and pathologists and can be confused with malignant uterine neoplasms. Only a few cases have been reported so far in the literature. Here we report a case of a cotyledonoid dissecting leiomyoma in a 60-year-old woman, emphasize its gross and histological features, and provide a review of the literature.

Keywords: Uterus; Smooth muscle; Cotyledonoid; Leiomyoma

Introduction

Leiomyomas are benign smooth muscle neoplasms that arise from the myometrium and account for almost 75% of hysterectomy cases [1]. Such cases are mostly seen in women of reproductive age with a low incidence in postmenopausal age. Leiomyomas have numerous morphologies, among which cotyledonoid dissecting leiomyoma (CDL) is a very unusual form. Its rarity and unfamiliarity may lead to its misdiagnosis as a malignant tumor [2]. Here we describe a case of CDL in a post-menopausal woman who presented with lower abdominal pain and third-degree uterine prolapse and present a review of the literature.

Case report

A 60-year-old woman presented with the complaint of a 1-year history of lower abdominal pain and third-degree uterovaginal descent. Her previous menstrual history was unremarkable. A vaginal examination revealed a bulky uterus with an ulceration on the anterior cervical lip. Ultrasonography revealed that the uterus was 7.5×3.4 cm in size with an endometrial thickness of 4.9 mm. Multiple uterine fibroids 2–4 cm in diameter were also noted. Both adenexa were unremarkable. A hysterectomy was performed and the specimen was sent for histopathological examination. Grossly, the cervix appeared hypertrophied and epidermidized. The endomyometrial thickness was 1.6 cm, while the endometrial thickness was 0.4 cm. Multiple fibroids up to 4×3.5 cm were noted. In addition, 2 subserosal fibroids 2 cm and 3.5 cm in diameter were seen. A cut section of the largest intramural fibroid and 1 subserosal fibroid revealed a solid, grayish-white, homogenous, and whorled appearance. The cut section of another subserosal fibroid revealed the presence of multiple grayish-white nodules (Fig. 1A). Microscopy revealed nodules of varying sizes of uniform smooth muscles arranged in interlacing and whorling fascicles with few prominent blood vessels (Fig. 1B–D). However, no signs of significant mitotic activity, nuclear atypia, or necrosis were seen. Based on the gross features and microscopic findings, a final diagnosis of CDL was made. Immunohistochemistry performed

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on CDL sections revealed diffuse positivity for vimentin, desmin, and smooth muscle actin, confirming the histopathological diagnosis of CDL (Fig. 2). In the present case, this rare variant of leiomyoma was associated with multiple intramural and subserosal classical leiomyomas showing features of hyaline degeneration (Fig. 3A). The endometrium was in the proliferative phase and adenomyosis was noted in the myometrium (Fig. 3B). The cervix showed features of acute and chronic cervicitis with surface ulceration and keratinization.

Discussion

Leiomyomas are the most common benign smooth muscle neoplasms of the uterus. A number of patterns of leiomyomas have been described. CDL, a very rare variant that is also

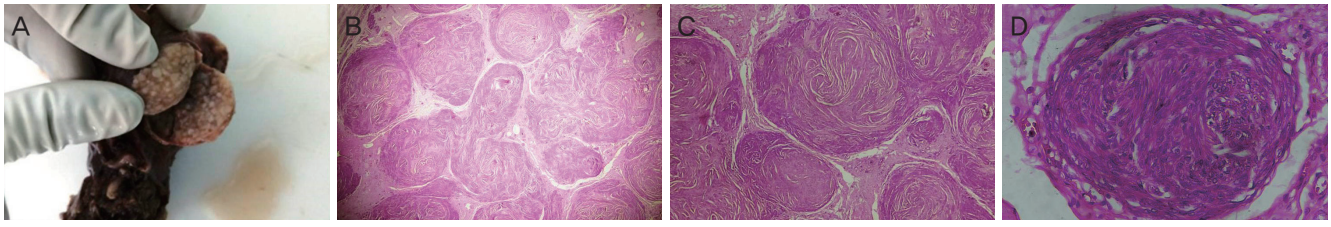

Fig. 1. Gross and histopathology images of cotyledonoid dissecting leiomyoma. (A) Cut section with multiple tan white nodules. (B) Classical whorling pattern (hematoxylin and eosin [HE], ×20). (C) Nodules of varying sizes of uniform smooth muscles arranged in interlacing and whorling fascicles with few prominent blood vessels (HE, ×100). (D) Bland smooth muscles arranged in an interlacing pattern with no signs of nuclear atypia, mitosis, or necrosis (HE, ×400).

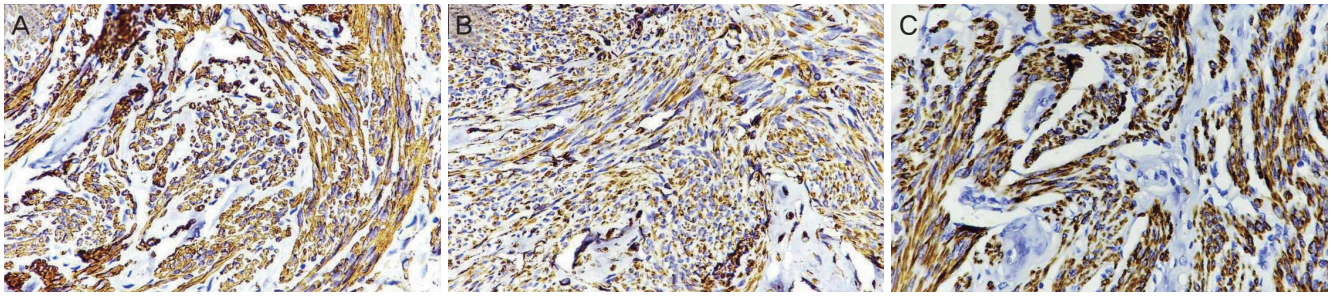

Fig. 2. Immunohistochemistry images of cotyledonoid dissecting leiomyoma showing: (A) smooth muscle actin (immunohistochemistry [IHC], ×400); (B) Vimentin (IHC, ×400); and (C) Desmin positivity in smooth muscle fibers (IHC, ×400).

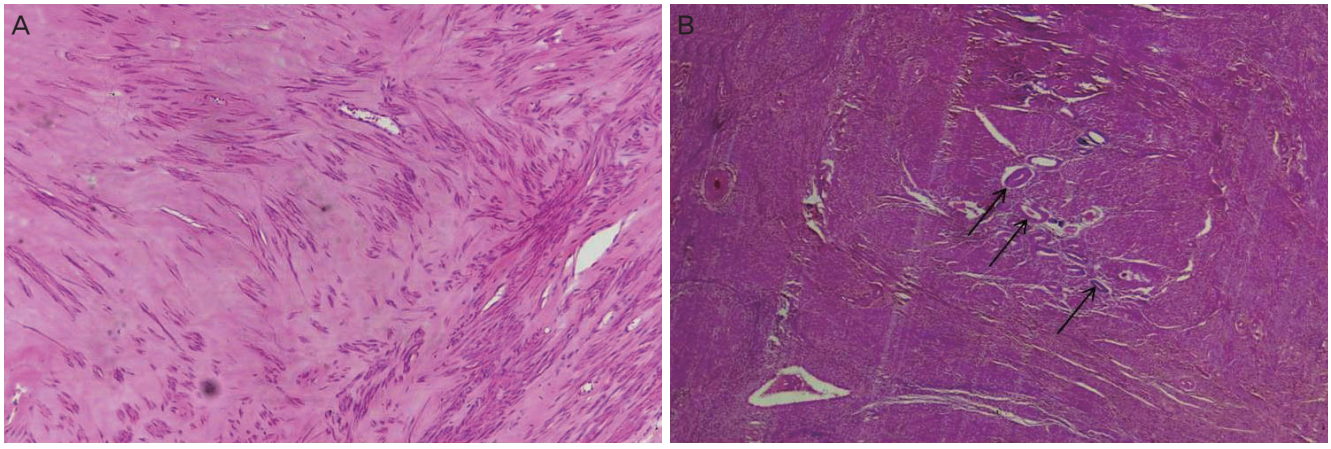

Fig. 3. Histopathological images of intramural leiomyoma showing: (A) Benign smooth muscles arranged in an interlacing pattern with large areas of hyalinization (hematoxylin and eosin [HE], ×100) and myometrium; and (B) Features of adenomyosis (HE, ×40).

Table 1. Summary of the published cases of cotyledonoid dissecting leiomyoma

SN	Reference	Age	Clinical presentation	Tumor size (cm) maximum dimension	Tumor location
1	David et al. [1]	65	Abnormal uterine bleed	15	Uterine fundus and cervix
		48	Uterine prolapse	12	Uterine fundus
2	Roth et al. [2]	39	Pelvic mass	10.3	Uterine cornua
		41	Abnormal uterine bleed	10	Uterine cornua
		23	Pelvic mass	25	Uterine cornua
		Unknown	Pelvic mass	24	Uterine cornua
3	Brand et al. [3]	24	Abdominal mass	NA	Uterine fundus
4	Roth and Reed [4]	46	Pelvic mass	34	Uterine cornua
5	Kim et al. [5]	26	Incidental	12	Posterior uterine wall
6	Cheuk et al. [6]	55	Abnormal uterine bleed	10	Uterine cornua
7	Stewart et al. [7]	58	Abdominopelvic mass	16.4	Uterine fundus
8	Jordan et al. [8]	46	Right adnexal mass	22	Uterine with extrauterine extension (all cases)
		46	Pelvic mass	20	
		46	Pelvic mass	10	
		46	Pelvic mass	18	
		36	Abnormal uterine bleed	13	
		34	Uterine mass, infertility	18	
9	Saeed et al. [9]	27	Pelvic mass	41	Uterine fundus
10	Maimoon et al. [10]	40	Urinary retention	10	Uterine isthmus
11	Shelekhova et al. [11]	73	Uterine mass	8	Uterine fundus
12	Gurbuz et al. [12]	67	Incidental	10	Uterine cornua
13	Weissferdt et al. [13]	52	Abnormal uterine bleed	6.2	Uterine fundus
14	Raga et al. [14]	33	Abdominal pain	6	Lateral part of uterus
15	Driss et al. [15]	47	Pelvic mass	25	Uterine with extrauterine extension
16	Preda et al. [16]	41	Uterine mass	9	Left and posterior uterine wall
17	Fukunaga et al. [17]	56	Constipation	30	Posterior uterine wall
		47	Abdominal pain	26	Posterior uterine wall
		36	Abnormal uterine bleed	4	Posterior uterine wall
		35	Abdominal pain	18	Lateral uterine wall
18	Gezginç et al. [18]	57	Pelvic pain	2.5, 4.5	Intrauterine, lateral uterine wall
19	Agarwal et al. [19]	52	Abnormal uterine bleeding	10	Uterine cornua
20	Ersöz et al. [20]	51	Abnormal uterine bleeding	8.5	Subserosal
21	Roth et al. [21]	33	Abnormal uterine bleeding	6.5, 13.5	Posterior uterine wall
22	Tanaka et al. [22]	36	Incidental	10	Posterior & lateral uterine wall
23	Onu et al. [23]	50	Incidental	10	Uterine fundus
24	Kim et al. [24]	43	Abdominal mass	13	Uterine with extrauterine extension
25	Blake et al. [25]	56	Abnormal uterine bleeding	30	Uterine with extrauterine extension
26	Shimizu et al. [26]	40	Abnormal uterine bleeding	10	Posterior uterine wall
27	Rocha et al. [27]	38	Abnormal uterine bleeding	25	Uterine isthmus
28	Xu et al. [28]	55	Pelvic mass	6	Posterior uterine wall
		43	Pelvic mass	3	Body of uterus
		37	Pelvic mass	30	Peri- uterine
		48	Lower abdominal pain	6.7	Right wall of uterus

commonly known as Sternberg tumor, was first reported by Roth et al. [3] Menolascino-Bratta et al. [4] coined the term "angionodular dissecting leiomyoma". These tumors are frequently seen in the third to sixth decades of life. The most common complaints are lower abdominal pain and abnormal uterine bleeding. The apex case also presented with complaints of lower abdominal pain; however, no vaginal bleeding was revealed. Tumor size was typically 2–15 cm [4,5]. Three types of CDL have been described in the literature. The first appears as an exophytic mass of multinodular tissue protruding from the lateral surface of the uterine cornua; resembling the placenta is called CDL. The second type is an intramural dissecting tumor that is confined to the uterus. These 2 types share similar histopathological features. The last type is pure cotyledonoid leiomyoma, which is not associated with either a parent intramural mass or intramural dissection [6]. This case met the criteria for exophytic CDL. Microscopically, it is characterized by nodules of various sizes of uniform smooth muscles arranged in interlacing and whorling fascicles. Many blood vessels are also prominent with focal hypercellular areas. However, in contrast to malignant lesions, signs of mitotic activity, nuclear atypia, cellular pleomorphism, and necrosis are absent. Vascular invasion, capsular infiltration, and metastasis are not seen. In a few cases, perinodular hydropic changes may be prominent [7].

A variety of other unusual patterns of uterine leiomyoma have been described, such as parasitic leiomyoma, cellular leiomyoma, symplastic or bizarre leiomyoma, epithelioid leiomyoma, intravenous leiomyomatosis, and leiomyoma with secondary changes. Some CDL appear as large fungating masses with widespread extension into the broad ligament and pelvic cavity. Due to its rarity and a clinician's lack of familiarity, such tumors are sometimes misdiagnosed as malignancies [8].

Gurbuz et al. [9] reported a case of cotyledonoid leiomyoma that had no intrauterine portion but had extrauterine extensions. A comparative analysis of various CDL cases reported in the literature is given in Table 1 and Supplementary Data 1.

In conclusion, CDL is a unique and rare variant of leiomyoma with a characteristic gross nodular appearance and microscopic features. Increasing awareness among clinicians and pathologists regarding this rare entity will prevent inappropriate diagnosis and treatment.

Conflict of interest

No potential conflict of interest relevant to this article was reported.

Ethical approval

The study was done in adherence to the Institutional Ethics committee guidelines (All India Institute of Medical Sciences, Patna, Bihar, India). As the study is purely based on tissue sample and slides which were used after routine reporting and no clinical/drug trial was done it was not registered in ethics committee as per Institutional Ethics committee guidelines, hence Institutional Review Board (IRB) number was not obtained. The study performed in accordance with the principles of the Declaration of Helsinki. Written informed consents were obtained.

Patient consent

The present work was performed after the patient provided informed consent, and a sincere effort has been made to uphold patient confidentiality.

Supplementary material

Supplementary Data 1 associated with this article can be found online at https://doi.org/10.5468/ogs.2019.62.5.362.

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