

A Case Report on Recurrent Cotyledonoid Dissecting Leiomyoma

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ABSTRACT Cotyledonoid dissecting leiomyoma is an extremely rare variant of uterine leiomyomas. Although they are pathologically benign, they manifest as sarcomatoids because of their appearance. A 34-year-old single patient was admitted to the Obstetrics and Gynaecology outpatient clinic for recurrent abnormal uterine bleeding and inguinal pain. The patient underwent hysterectomy with a diagnosis of recurrent cotyledonoid dissecting leiomyoma. In this report, we discuss the aggressive clinical course of a patient with recurrent cotyledonoid dissecting leiomyoma resulting in hysterectomy, which has been reported in a limited number of studies in the literature.

Keywords: Leiomyoma; cotyledonoid; dissecting; Stenberg tumour; leiomyosarcoma

Leiomyomas are benign pelvic tumours originating from smooth muscle cells of the myometrium and are most commonly observed in women.^{1,2} Hendrickson and Kempson divided primary benign smooth muscle tumours into three categories: leiomyomas with usual differentiation, variants defined on the basis of cytological features or cellularity and those with unusual patterns of growth.³

Cotyledonoid dissecting leiomyoma, dissecting leiomyoma and cotyledonoid leiomyoma are the rare variants of leiomyoma. Cotyledonoid dissecting leiomyoma, defined by Roth et al. in 1996, is a rare variant of uterine leiomyomas. The same study also named it as Stenberg tumour, in dedication to the contributions of William H. Stenberg.⁴ Previously, unusual uterine leiomyomas exhibiting exophytic growth pattern were defined as 'grapelike leiomyoma' in 1975 and 1980.⁵⁻⁷

The name 'cotyledonoid' has been used because of the similarity of the extrauterine component of the mass to cotyledons adhering to the interior sections of the uterine surface of the placenta. It was defined as a cotyledonoid dissecting leiomyoma because the exophytic components of the mass have continuity with intramural components. Intramural components divide the myometrium into fascicles in an irregular pattern.⁴ In laparotomy, an exophytic tumour extending from the lateral surface of the anterior side of the uterus to the pelvic space is observed. It is noteworthy that there is no adhesion between the exophytic part of the cotyledonoid dissecting leiomyoma and normal adjacent tissues. Although cotyledonoid dissecting

leiomyomas exhibit a sarcomatoid appearance owing to their unusual macroscopic appearance, they are histologically benign owing to the absence of cellular atypia, mitotic activity and coagulative tumour necrosis.⁴

A study conducted in 1999 reported eight cases of only intramural dissection, and another study conducted in 2000 reported a case of only cotyledonoid leiomyoma without presentation of any dissecting form.^{8,9}

CASE REPORT

A 34-year-old single patient presented at our clinic with suprapubic pain and irregular and excessive menstrual bleeding. The patient had a history of three myomectomies with one laparotomy and one hysteroscopy. Family history revealed that the patient's sister had two myomectomies. The patient's pathologies for myomectomies indicated cotyledonoid dissecting leiomyoma.

Pelvic examination revealed that the uterus size was equal to the size of a 3-month pregnant women's uterus, and masses of various sizes were identified in the uterus. Pelvic ultrasonography revealed a large number of pelvic masses of various sizes, with the largest being 6 cm. Both the ovaries had normal appearance. Tumour markers were within normal ranges. Surgical treatment was decided owing to the presence of excessive vaginal bleeding, severe inguinal pain that increased during the menstrual period and recurrent leiomyoma despite previous treatment.

The patient's abdomen was opened with an 8-cm midline incision and exploration revealed two cotyledonoid dissecting leiomyomas on the anterior side of the uterus (masses of 17×11 and 12×6 cm near the right- and left-side walls, respectively); a 10×10-cm subserous myoma in the left cornual region; a 4×4-cm subserous myoma in the right cornual region and several intramural and subserous myomas of various sizes (Figure 1). Both ovaries had normal appearance. Invasion of cotyledonoid masses in the surrounding tissues was not observed. During surgery, the frozen procedure was performed owing to the sarcomatous appearance of the masses. How-

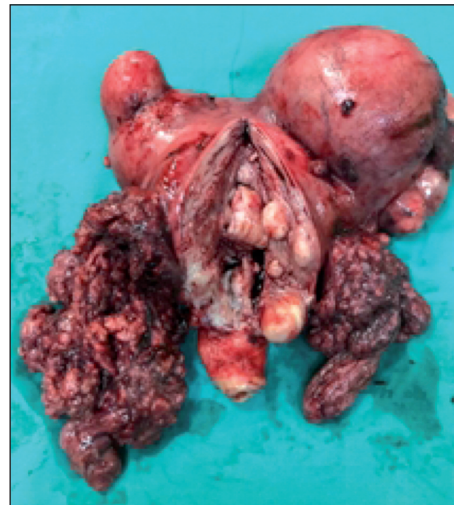


FIGURE 1: Exploration revealed two cotyledonoid dissecting leiomyomas on the anterior side of the uterus (masses of 17×11- and 12×6-cm near the right and left side walls respectively); a 10×10-cm subserous myoma in the left cornual region; a 4×4-cm subserous myoma in the right cornual region and several intramural myomas of various sizes.

ever, the frozen procedure failed to differentiate between benign and malignant masses. The patient was informed regarding the possibility of sarcomatous transformation. Total abdominal hysterectomy (TAH) was performed because of the history of recurrent deep anaemia; presence of cotyledonoid dissecting leiomyomas with atypical appearance and large size on the anterior wall of the uterus; the large number of recurrent subserous, intramural and submucous myomas of various sizes and chronic pelvic pain. The pathology report of the patient indicated a cotyledonoid dissecting leiomyoma. The patient's was discharged with good health 2 days after the surgery.

DISCUSSION

Uterine smooth muscle tumours exhibit a wide range of growth patterns. Cotyledonoid dissecting leiomyomas have been observed in patients aged 23-65 (mean 40.3) years, depicting a wide range of age. Pelvic mass and abnormal uterine bleeding are the most common clinical presentations, similar to those observed in the patient in our study. These tumours are generally large, with an average dimension of 17.7 cm.¹⁰ These tumours mainly emerge from the fundus or the posterior aspect of

the cornu of the uterus. The differential diagnosis of cotyledonoid leiomyomas includes intravenous leiomyomatosis, leiomyomas with perinodular hydropic changes and myxoid leiomyosarcoma. Although myxoid leiomyosarcomas demonstrate malignant biological nature, they do not have atypical cytology; moreover, their mitotic activity is low at 0–2 counts per 10 high-power fields.

In a study conducted in 2013, TAH was performed in a patient with a history of myomectomy with cotyledonoid dissecting leiomyoma owing to recurrence of the lesion after 5 years. This recurrence was possibly caused by insufficiency of the previous myomectomy.¹¹ However, in the present study, recurrent myomas after a history of myomectomy were attributed to the susceptibility of cotyledonoid dissecting leiomyoma for recurrence despite its benign histological nature rather than insufficiency of the previous surgery.

In conclusion, cotyledonoid dissecting leiomyomas are rare, with a malignant macroscopic appearance despite their benign histological nature. Even if they are pathologically benign, they can present with recurrent masses owing to their susceptibility for recurrence. The present study aimed to create awareness regarding cotyledonoid dissecting leiomyoma variants, which are characterised by

benign pathologies despite their sarcomatous appearance, among gynaecologists and pathologists.

In case the selected course of treatment is myomectomy, the study suggests that the patients should be closely monitored for possible recurrences, and planned pregnancies should be completed as soon as possible.

Source of Finance

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: Özgür Özdemir; **Design:** Özgür Özdemir; **Control/Supervision:** Özgür Özdemir; **Data Collection and/or Processing:** Özgür Özdemir; **Analysis and/or Interpretation:** Süleyman Güven; **Literature Review:** Gülten Sağır; **Writing the Article:** Özgür Özdemir; **Critical Review:** Abdulkadir Reis; **References and Fundings:** Bakiye Akbaş; **Materials:** Özgür Özdemir.

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