J Cases Obstet Gynecol, 2015;2(4):91-93

Case Report

Cotyledonoid myoma: a distinct entity and a diagnostic dilemma in gynecology

Sevda Bas[1], Ilker Selcuk[1,*], Levent Sirvan[2], Hakan Yalcin[1], Tayfun Gungor[1,3]

[1] Gynecologic Oncology Unit, Zekai Tahir Burak Women's Health, Education and Research Hospital, Ankara, Turkey
[2] Pathology Unit, Zekai Tahir Burak Women's Health, Education and Research Hospital, Ankara, Turkey
[3] Department of Obstetrics and Gynecology, Hitit University Faculty of Medicine, Corum, Turkey

Abstract

Leiomyomas are benign uterine tumors and they are the most common tumor of female genital tract. 'Cotyledonoid myoma', also known as 'Sternberg tumor', is a rare type of leiomyoma and has a unique nature with surgery as the main step in its treatment. The differential diagnosis of cotyledonoid myoma from a malignant condition is important for the follow up of patients. Here we present the clinical evaluation of a 46 years-old patient with cotyledonoid myoma.

Key words:

Cotyledonoid, myoma, gynecology, hysterectomy, sarcoma

Introduction

Leiomyomas are benign uterine tumors mostly seen in reproductive aged women and are the most common tumor of female genital tract [1]. Generally they have the characteristic view of a well-circumscribed round, pearly white, firm tissue. Nevertheless some distinct myoma types may grow and disseminate discretely. Roth et al. [2] in 1996 defined an unusual type of myoma as "cotyledonoid myoma". Cotyledonoid myoma has a dissecting nature towards the myometrium and also has a reddish, exophytic, placenta like gross appearance with the usual extension into the broad ligament and pelvic cavity. Additionally perinodular hydropic degeneration is a common characteristic of these tumors [3, 4]. Despite these features cotyledonoid tumors are benign in nature [5]. In this report we describe a case of cotyledonoid myoma.

Article history:
Received 02 02 2015
Accepted 18 07 2015

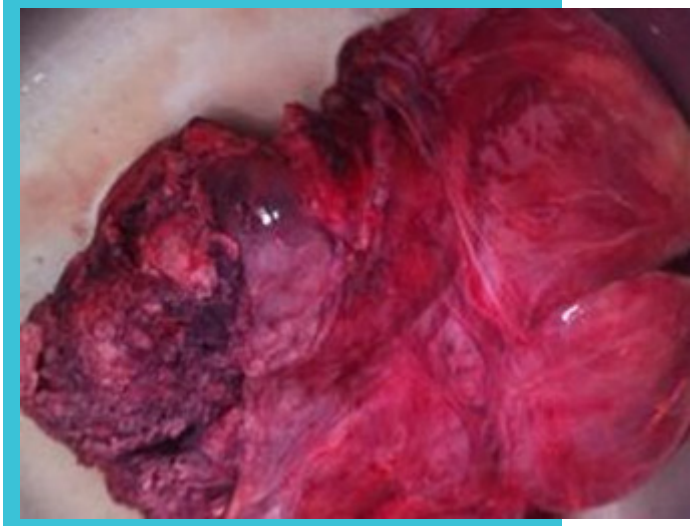
*** Correspondence:** Ilker Selcuk
Gynecologic Oncology Unit, Zekai Tahir Burak Women's Health, Education and Research Hospital, 06030, Altindag, Ankara, Turkey.
Phone: 090 312 306 50 00
Email: ilkerselcukmd@hotmail.com

Case presentation

A 46 years-old previously healthy woman, gravidity 2 para 2 had presented with pelvic pain and menometrorrhagia to an outside hospital. Her symptoms had been terrifying her since the last six months. She had been operated because of the suspicion of a myoma at the outside hospital. The gynecologist could not resect the full mass thus a partial resection had been performed and the patient was referred to our center; Dr. Zekai Tahir Burak Women's Healh, Education and Research Hospital. We performed an abdominal computed tomography and a 25x18x15 cm solid-cystic not well- circumscribed contrast filled mass which was causing compression on rectum and bladder was observed. The tumor markers were in normal limits. We planned an explorative laparotomy. Intraopera-

tively a gross, multinodular, thick, irregular bordered mass with a right sided deviation; originating from the uterus lying towards the douglas posteriorly and extending to the outside of pelvis superiorly was detected. Although the mass gave the impression of a sarcoma at first sight, the frozen section analysis defined the mass as a benign smooth muscle tumor.

Figure 1.



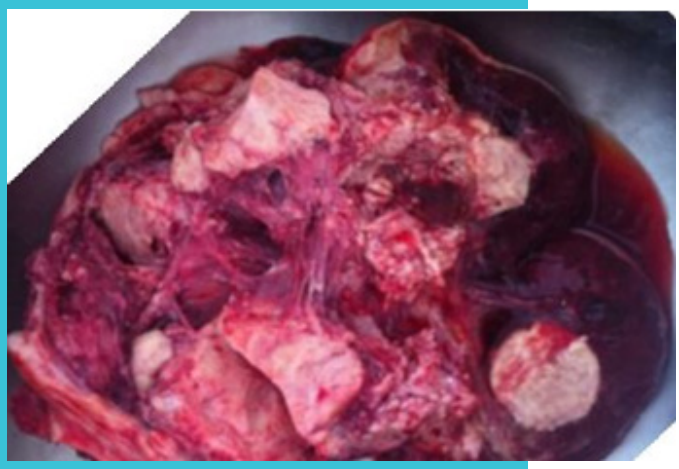
Red-brown, solid, multinodular, shaggy mass

The uterus was resected completely and the final pathology result revealed cotyledonoid myoma. The patient was well after the operation. No lesion was detected by abdominal sonographic evaluation six months after operation. The patient was put on a clinical follow-up schedule with pelvic examination and abdomino-pelvic ultrasound yearly. The specimen was irregularly bordered and had a multinodular, red-brown, craggy-solid macroscopy with dimensions of 22x16x15 cm and it was 2.3 kg in weight. Smooth to shaggy surface areas and a degenerative view established the initial work-up. (Figure 1 and 2) Microscopically whorling fascicles of uniform smooth muscle tumor without atypia, mitosis and coagulative necrosis was seen. The tumor was originating from the interface between the myometrium and serosa. Edema and perinodular hydropic degeneration were present with increased vascularity nevertheless no intravascular growth was detected.

Discussion

In 1975 David et al. [6] reported two cases with grape-like leiomyomas of uterus. However in 1996 Roth et al. [2] defined a different variant of this entity in four cases and called it "cotyledonoid dissecting leiomyoma" due to its gross resemblance to placenta. It is also known as "Sternberg tumor". Although the tumor has some characteristic features such as a bulky, reddish and exophytic, grape-like mass and a benign histologic nature without atypia and coagulative necrosis [7], its pre-operative sonographic view may mimic a malignant condition. It is frequently seen as an irregular bordered, bulky, solid-cystic mass. The tumor may have the shape of a solid mass at the pelvic cavity, descending towards the Douglas pouch, rectum and pressing upwards to the bladder and gastrointestinal tract. The mass will extend towards the adnexal areas and gastrointestinal region which may give the impression of an ovarian mass or a gastrointestinal tumor with symptoms of adjacent organ pathologies [4]. Pre- and intra-operative multidisciplinary approach is needed for this type of lesion. It may also mimic an ovarian tumor.

Figure 2.



Posterior aspect of giant mass with nodular grappy appearance

In the literature a case of cotyledonoid tumor was reported and it was followed by clinicians as a persisting ovarian cyst [7]. Intraoperative findings show that cotyledonoid leiomyoma frequently originates from the subserosal myometrium of the lateral parts of uterus and grows exophytically [3].The morphologic appearance can be mistaken for a sarcoma. In this setting contrast-enhanced dynamic magnetic resonance imaging (MRI) may evaluate the origin of the lesion and may be useful in differential diagnosis [4, 8]. Although Roth et al. [2] identified the patients' age as ranging between 23-41; Gurbuz et al. [7] reported a case of cotyledonoid leiomyoma for a 67 years-old patient. However, most patients are in reproductive ages. Abnormal uterine bleeding and a pelvic mass with pain are the leading symptoms. Nevertheless uterine prolapsus [9] or rectal-urinary symptoms sometimes bleeding or incontinence [10] may be the initial symptoms. The case presented here was 46 years-old and admitted with the typical symptoms of pelvic mass and abnormal uterine bleeding. Sonographically a bizarre shaped, bulky and solid mass firstly gave the impression of a pelvic lesion at the outside hospital. Afterwards intraoperatively a reddish-brown, exophytic, gross, multinodular solid mass, like the identified cases in the literature aroused our suspicion for a malignant neoplasm as a leiomyosarcoma. Intravenous leiomyomatosis, leiomyoma with perinodular hydropic degeneration, low-grade endometrial stromal sarcoma and leiomyosarcoma should be differentiated from cotyledonoid dissecting leiomyoma. Immunohistochemistry, findings of atypia, mitosis or coagulative necrosis are paramount features in differential diagnosis [4]. Cotyledonoid dissecting leiomyomas are benign tumors without atypia, mitosis and coagulative necrosis. They do not recur. However, after incomplete resection the tumor may undergo further growth [11]. In conclusion, cotyledonoid leiomyoma of the uterus is a very rare benign entity. Although the tumor has some characteristic views macroscopically that are not unique; microscopically subserosal growth, dissecting growth and perinodular hydrophic degeneration are important in diagnosis besides the lacking features of atypia, mitosis and necrosis. The most important aspect of this tumor is to be assumed as a malignant neoplasm preoperatively and intraoperatively.

Acknowledgments

This case report was presented as a poster presentation at International Gynecologic Cancer Society Congress in Melbourne, 2014.

Conflict of Interest

Authors declare no conflict of interest

References

1. Ciavattini A, Di Giuseppe J, Stortoni P, Montik N, Giannubilo SR, Litta P, et al. Uterine Fibroids: Pathogenesis and Interactions with Endometrium and Endomyometrial Junction. Obstet Gynecology Int. 2013:173184.
2. Roth LM, Reed RJ, Sternberg WH. Cotyledonoid dissecting leiomyoma of the uterus. The Sternberg tumor. Am J Surgical Pathol. 1996;20:1455-61.
3. Weissferdt A, Maheshwari MB, Downey GP, Rollason TP, Ganesan R. Cotyledonoid dissecting leiomyoma of the uterus: a case report. Diagnc Pathol. 2007;2:18.
4. Smith CC, Gold MA, Wile G, Fadare O. Cotyledonoid dissecting leiomyoma of the uterus: a review of clinical, pathological, and radiological features. Int J Surg Pathol. 2012;20:330-41.
5. Kim MJ, Park YK, Cho JH. Cotyledonoid dissecting leiomyoma of the uterus: a case report and review of the literature. J Korean Med Sci. 2002;17:840-4.
6. David MP, Homonnai TZ, Deligdish L, Loewenthal M. Grape-like leiomyomas of the uterus. Int Surg. 1975;60:238-9.
7. Gurbuz A, Karateke A, Kabaca C, Arik H, Bilgic R. A case of cotyledonoid leiomyoma and review of the literature. Int J Gynecol Cancer. 2005;15:1218-21.
8. 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.
9. Cheuk W, Chan JK, Liu JY. Cotyledonoid leiomyoma: a benign uterine tumor with alarming gross appearance. Arch Pathol Lab Med. 2002;126:210-3.
10. Stewart KA, Ireland-Jenkin K, Quinn M, Armes JE. Cotyledonoid dissecting leiomyoma. Pathology. 2003;35:177-9.
11. Roth LM, Kirker JA, Insull M, Whittaker J. Recurrent cotyledonoid dissecting leiomyoma of the uterus. Int J Gynecol Pathol. 2013;32:215-20.