Case Report

Atypical cell leiomyoma of the uterus with amianthoid-like fibers: A case report.

Longo F1, Musumeci G2*, Parenti R3, Vecchio G1, Magro G1

1Department G.F. Ingrassia, Azienda Ospedaliero-Universitaria “Policlinico-Vittorio Emanuele” Anatomic Pathology, University of Catania, Catania, Italy

2Department of Bio-Medical Sciences, Human Anatomy and Histology Division, University of Catania, Italy.

3Department of Bio-medical Sciences, Section of Physiology, University of Catania, Italy.

*Corresponding author
E-mail: g.musumeci@unict.it.

Manuscript type: Case Report
Sections: Anatomy and Anatomic Pathology
All authors contributed to conception and design, manuscript preparation, read and approved the final manuscript.
All authors abide by the Association for Medical Ethics (AME) ethical rules of disclosure.
Competing interests: none declared, Conflict of interests: none declared.
Abstract

Amianthoid-like fibers are hyalinized collagen mats which are characteristically found in intranodal palisaded myofibroblastoma. Only rarely they can be encountered in uterine classic-type leiomyomas. We herein report the first case of uterine atypical/bizarre cell leiomyoma which contained, as unusual and striking feature, numerous amianthoid-like collagen fibers. Diagnostic and histogenetic considerations are discussed. Pathologist should be aware of the possibility that the most diagnostically challenging variant of uterine leiomyoma, namely atypical/bizarre cell leiomyoma, may contain amianthoid-like fibers to avoid confusion with other tumors.

Key words: Amianthoid-like fibers; myofibroblastoma; uterine leiomyoma.
Introduction

Uterine leiomyoma is a benign smooth muscle neoplasm, typically arising in the uterus, and more rarely in deep soft tissues. Uterine leiomyoma occurs as a single or multiple circumscribed nodules, especially in women over the age of 50 years. Metrorrhagia is the most frequent symptom especially in submucosal-located tumours. Grossly, uterine leiomyoma appears as a well-circumscribed, whitish nodule, which is histologically composed of interlacing fascicles of bland-looking spindle-shaped cells, with abundant, deeply eosinophilic cytoplasm and elongated nuclei with blunt ends (so-called “cigar-shaped”). Over the last decades, a wide variety of histologic patterns has been recognized in uterine leiomyoma: i) cellular, ii) mitotically active, iii) atypical/bizarre cell, iv) epithelioid (clear cell), v) lipomatous (so called “lipo-leiomyoma”), vi) vascular (so-called “angioleiomyoma”), and vii) cotyledonoid/dissecting variants (1). In addition the following morphologic features have been described in the context of an otherwise typical leiomyoma: i) hydropic/myxoid changes; ii) granular cell changes; iii) rhabdomyoblastic-like changes; iv) palisaded growth pattern; v) coagulative necrosis; and vi) diffuse lymphoid infiltration1. Although the accurate classification of uterine leiomyomas may seem to be of academic interest, the potential impact of recognizing their more unusual variants, especially atypical/bizarre cell leiomyoma with diffuse nuclear pleomorphism, is crucial to avoid a misdiagnosis of malignancy.

Only rarely uterine leiomyomas may contain deeply eosinophilic thick collagen bands, also known with the term of “amianthoid-like fibers”. 2,3 To the best of our knowledge we report the first case of an atypical/bizarre cell uterine leiomyoma with amianthoid-like fibers. Clinico-pathological features and differential diagnostic problems are discussed.

Case Report

A 45-year old woman was referred to our hospital for laparascopic myomectomy. Five subserosal leiomyomas, ranging in size from 1 to 5 cm in greatest diameter, were excised. For routine
histology, surgically obtained samples were fixed in 4% buffered formalin, and the specimens were embedded in paraffin. Sections (5µm) were stained with hematoxylin and eosin (H&E). Additional sections were cut for histochemical (van Gieson, Goldner trichrome, Gomori and Congo red staining) and immunohistochemical procedures (Data not shown). Immunohistochemical studies were performed with the labeled streptavidin-biotin peroxidase detection system using the Ventana automated immunostainer (Ventana Medical Systems, Tucson, AZ). The antibodies tested were α-smooth muscle actin, desmin, h-caldesmon, S-100 protein, CD34, CD10, cytokeratins (all from DakoCytomation, Glostrup, Denmark), (Data not shown).

Grossly, all nodules were well-circumscribed, whitish in colour and showed whorled cut surface. Histologically all but one lesion showed the features of the classic-type leiomyoma. Conversely the tumor of largest size (5 cm in greatest diameter) showed morphological features consistent with an “atypical/bizarre cell leiomyoma”. Apart from the typical features of leiomyoma, this tumor showed focal areas containing mono- or multi-nucleated cells with mild to moderate degree of nuclear pleomorphism (Fig. 1). Only rarely mitoses were found (<1 mitosis x 10 HPF). Necrosis was absent. Notably the most striking morphological feature of this tumor was the presence of numerous round- to stellate-shaped, deeply eosinophilic, collagenous mats closely reminiscent of “amianthoid-like fibers” (Fig. 2-4). These fibers were also stained with van Gieson, Goldner trichrome, and Gomori staining, while were negative for Congo red staining (Data not shown).

Similar “amianthoid-like fibers” have originally been described in intranodal palisaded myofibroblastoma,\(^4,5\) meningioma\(^6\) and in rare cases of benign soft tissues tumors.\(^7\) Immunohistochemically the neoplastic cells, including the atypical/bizarre cells, showed a diffuse staining for desmin, α-smooth muscle actin and h-caldesmon, while no staining was obtained for CD10, S-100 protein and cytokeratins (Data not shown). Based on morphological and immunohistochemical features, the diagnosis of “atypical/bizarre cell leiomyoma with amianthoid-like fibers” was rendered.
Discussion

Although the recognition of uterine leiomyoma by pathologists is straightforward in most cases, establishing a correct diagnosis may be difficult if one is dealing with an unusual variant, especially atypical/bizarre cell leiomyoma. In the present case, although the morphological appearance was characteristic of an “uterine atypical/bizarre cell leiomyoma”, it was the co-existence of numerous amianthoid-like fibers that caused some diagnostic problems. To the best of our knowledge, this is the first case of “uterine atypical/bizarre cell leiomyoma” containing numerous amianthoid-like fibers. These collagen fibers may be encountered in both non-neoplastic or neoplastic lesions, but they are characteristically detected in “intranodal palisaded myofibroblastoma”. Only rarely they have been described in meningiomas and “polypoid angiomyofibroblastoma-like tumor” of the oral cavity. Only three cases of uterine classic-type leiomyomas with amianthoid-like fibers have been reported in the English literature so far: the first case described by Fisher & El-Bahrawy in 2008, and two additional cases, respectively a uterine leiomyoma and an extra-uterine pelvic leiomyoma, reported by Zamencik and Kascák in 2011.

Although in our case the identification of amianthoid-like fibers was an unexpected finding, however both morphological and immunohistochemical features were consistent with an atypical/bizarre cell leiomyoma. In this regard neoplastic cells were arranged in interlacing fascicles and showed the typical features of mature smooth muscle cells. In addition some neoplastic cells exhibited a mild to moderate degree of nuclear pleomorphism (so-called “atypical or bizarre cells”). The smooth muscle nature of neoplastic cells was confirmed by diffuse coexpression of desmin, α-smooth muscle actin and h-caldesmon. The pathogenesis of the amianthoid-like fibers in uterine leiomyomas is still to be established. Some authors think that they may represent a degenerative phenomenon of pre-existing collagen fibers, while others state that these fibers may be the result from an active process of collagen secretion and deposition.

Apart from these pathogenetic hypotheses, the present paper contributes to widen the morphological spectrum of atypical/bizarre cell leiomyoma, the most diagnostically challenging
variant of uterine leiomyoma. Awareness of the possibility that uterine leiomyomas, including the atypical/bizarre cell variant, may contain numerous amianthoid-like fibers is helpful for pathologists to avoid confusion with other tumors. As these unusual collagen fibers have never been described in uterine leiomyosarcoma, their detection in an uterine smooth muscle tumor may be a useful marker of benignity.

**Consent**
Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editor-in-chief of this journal.

**Acknowledgement**
No financial assistance or support was received in any form for the review and writing this case report.

**Competing interests**
The authors declare that they have no competing interests.

**References**


Figure Legends

Fig. 1
This benign smooth muscle tumor showed areas containing cells with mild to moderate degree of nuclear pleomorphism (atypical or bizarre cells). This latter feature is consistent with the diagnosis of “atypical/bizarre cell leiomyoma” (H&E staining, magnification x100).
Fig. 2
Low magnification showing numerous thick collagen fibers admixed with smooth muscle cells (H&E staining, magnification x60).
Fig. 3
At higher magnification the collagen mats are consistent with “amianthoid-like fibers”: stellate shape and thread-like extensions, closely reminiscent of asbestos fibers, are seen (H&E staining, magnification x100).
Fig. 4
At higher magnification the collagen mats are consistent with “amianthoid-like fibers”: thread-like extensions, closely reminiscent of asbestos fibers, are seen (H&E staining, magnification x150).