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Atypical cell leiomyoma of the uterus with amianthoid-like fibres: a case report

F Longo¹, G Musumeci^{2*}, R Parenti³, G Vecchio¹, G Magro¹

Abstract

Introduction

Amianthoid-like fibres are hyalinised collagen mats which are characteristically found in intranodal palisaded myofibroblastoma. Only rarely can they be encountered in uterine classic-type leiomyomas. We herein report the first case of uterine atypical/bizarre cell leiomyoma, which contained an unusual and striking feature—numerous amianthoid-like collagen fibres. Diagnostic and histogenetic considerations are discussed.

Case report

Conclusion

The 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 fibres to avoid confusion with other tumours.

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 (“cigar-shaped”). Over the last decades, a wide variety of histologic patterns have been recognised in uterine leiomyoma: (i) cellular, (ii) mitotically active, (iii) atypical/bizarre cell, (iv) epithelioid (clear cell), (v) lipomatous (also called “lipo-leiomyoma”), (vi) vascular (also

called “angioliomyoma”), and (vii) cotyledonoid/dissecting variants¹. In addition, the following morphologic features have been described in the context of an otherwise typical leiomyoma: (i) hydrophic/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 recognising their more unusual variants, especially atypical/bizarre cell leiomyoma with diffuse nuclear

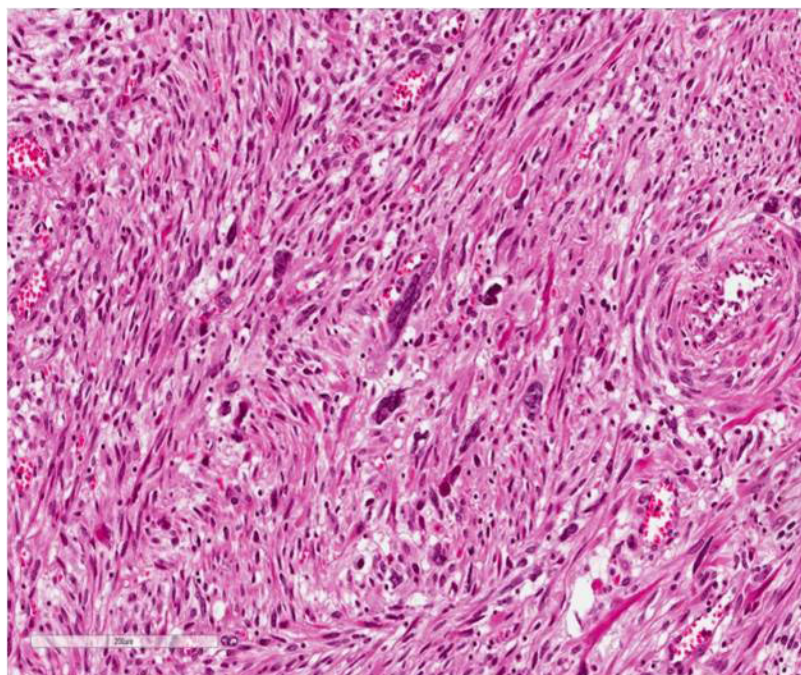


Figure 1: This benign smooth muscle tumour showed areas containing cells with mild to moderate degree of nuclear pleomorphism (atypical or bizarre cells). This latter feature is consistent with the diagnosis of “atypical/bizarre cell leiomyoma” (H&E staining, magnification $\times 100$).

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Case report

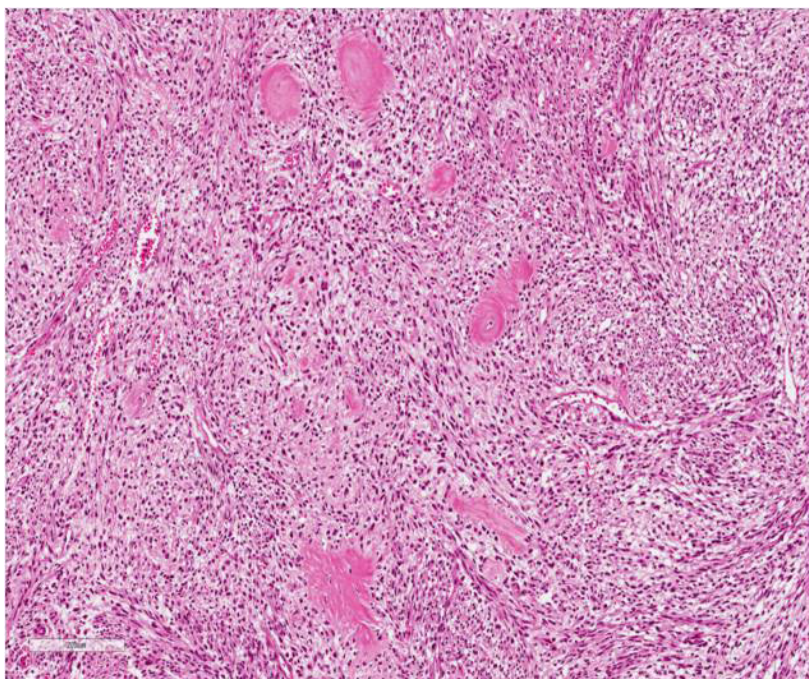


Figure 2: Low magnification showing numerous thick collagen fibres admixed with smooth muscle cells (H&E staining, magnification $\times 60$).

(van Gieson, Goldner trichrome, Gomori, and Congo red staining) and immunohistochemical procedures (data not shown). Immunohistochemical studies were performed with the labelled 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 tumour 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,

pleomorphism, is crucial to avoid a misdiagnosis of malignancy.

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

Case report

A 45-year old woman was referred to our hospital for laparoscopic 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 (5 μ m) were stained with haematoxylin and eosin (H&E). Additional sections were cut for histochemical

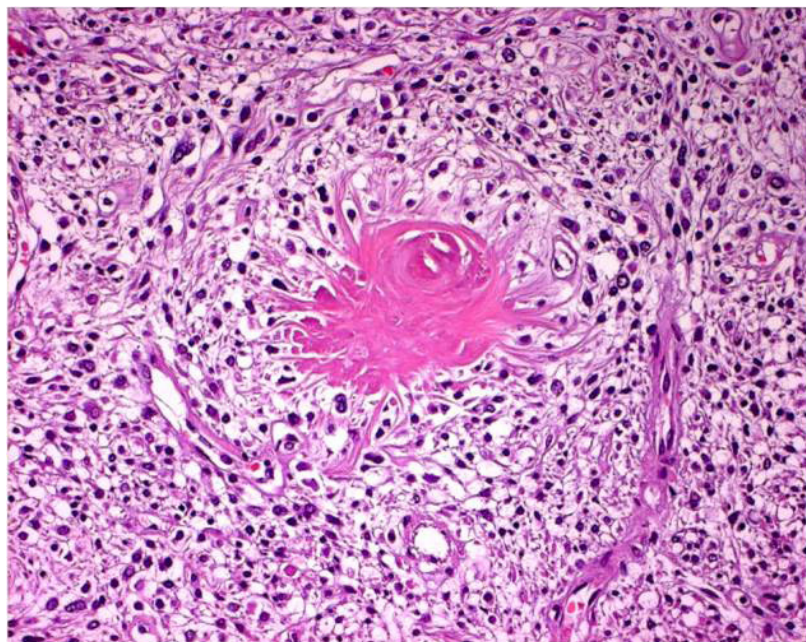


Figure 3: At higher magnification the collagen mats are consistent with “amiantoid-like fibres”: stellate shape and thread-like extensions, closely reminiscent of asbestos fibres, are seen (H&E staining, magnification $\times 100$).

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this tumour showed focal areas containing mono- or multi-nucleated cells with mild to moderate degree of nuclear pleomorphism (Figure 1). Only rarely mitoses were found (< 1 mitosis $\times 10$ HPF). Necrosis was absent. Notably the most striking morphological feature of this tumour was the presence of numerous round- to stellate-shaped, deeply eosinophilic, collagenous mats closely reminiscent of “amiantoid-like fibres” (Figures 2–4). These fibres were also stained with van Gieson, Goldner trichrome, and Gomori staining, while they were negative for Congo red staining (data not shown). Similar “amiantoid-like fibres” have originally been described in intranodal palisaded myofibroblastoma^{4,5}, meningioma⁶, and in rare cases of benign soft tissues tumours⁷. 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 amiantoid-like fibres” 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 a “uterine atypical/bizarre cell leiomyoma”, it was the co-existence of numerous amiantoid-like fibres that caused some diagnostic problems. To the best of our knowledge, this is the first case of “uterine atypical/bizarre cell leiomyoma” containing numerous amiantoid-like fibres. These collagen fibres may be encountered in both non-neoplastic and neoplastic lesions⁸, but they are

characteristically detected in “intranodal palisaded myofibroblastoma”^{4,5}. They have rarely been described in meningiomas⁶ and “polypoid angio-myofibroblastoma-like tumour” of the oral cavity⁷. Only three cases of uterine classic-type leiomyomas with amiantoid-like fibres have been reported in the literature so far: the first case described by Fisher and El-Bahrawy², and two additional cases, a uterine leiomyoma and an extra-uterine pelvic leiomyoma, reported by Zamencik and Kascák respectively³.

Although in our case the identification of amiantoid-like fibres was an unexpected finding, 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 (“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 amiantoid-like fibres in uterine leiomyomas is still to be established. Some authors think that they may represent a degenerative phenomenon of pre-existing collagen fibres⁹, while others state that these fibres may be the result of an active process of collagen secretion and deposition^{10,11}.

Conclusion

Apart from these pathogenetic hypotheses, this 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 amiantoid-like

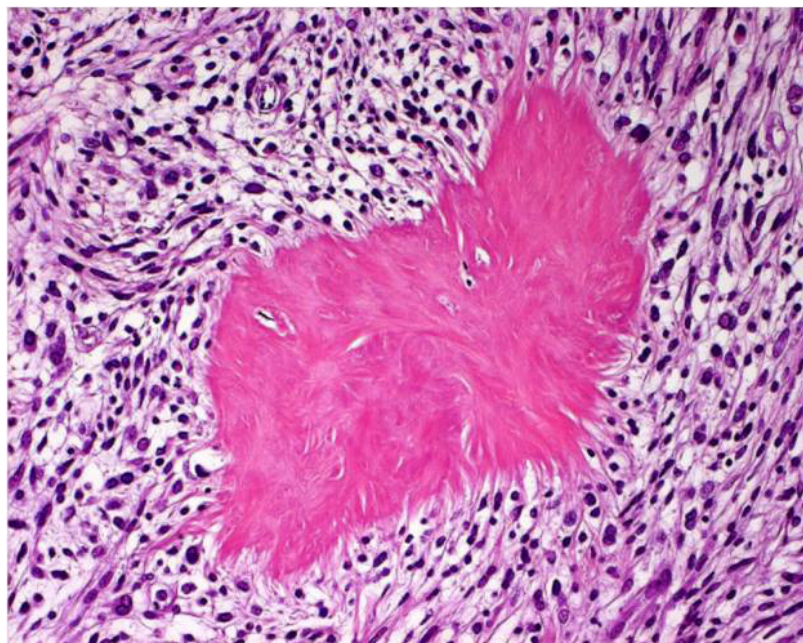


Figure 4: At higher magnification, the collagen mats are consistent with “amiantoid-like fibres”: thread-like extensions, closely reminiscent of asbestos fibres, are seen (H&E staining, magnification $\times 150$).

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fibres is helpful for pathologists to avoid confusion with other tumours. As these unusual collagen fibres have never been described in uterine leiomyosarcoma, their detection in a uterine smooth muscle tumour may be a useful marker of benignity.

Abbreviations list

H&E, haematoxylin and eosin.

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

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