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Uterine adenofibroma: Case report and literature review

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Abstract: *Uterine adenofibroma is an extremely rare benign tumour that occur, preferentially, in peri and post-menopausal women. It present both clinically and radiologically as benign tumours. In some cases, patients may be asymptomatic with a physical examination may be entirely unremarkable. On ultrasound examination, uterine adenofibromas perfectly simulates an endometrial polyp or a submucosal leiomyoma. As clinical and sonographic features are non-suggestive, the pathological examination remains the gold standard for the diagnosis, revealing a biphasic epithelial and mesenchymal tumour limited to the endometrium or cervical mucosa. Histologically, uterine adenofibroma must be particularly differentiated from adenosarcomas. In this paper, we report a case of an endometrial adenofibroma with a brief literature review. The aim of this work is to remind practitioners of the importance of the anatomopathological study, being the gold standard in the diagnosis of uterine adenofibromas, and to insist on hysterectomy as the treatment of choice for this tumour given its potential for recurrence, invasion and metastasis, as well as the possibility of coexistence with adenosarcoma. However, in young patients wishing to conceive, hysteroscopic lumpectomy may be a therapeutic alternative with a long-term follow-up.*

Keywords: *Uterine adenofibroma, adenosarcoma, endometrial tumour, rare, pathology, biphasic tumour, hysterectomy, hysteroscopic lumpectomy.*

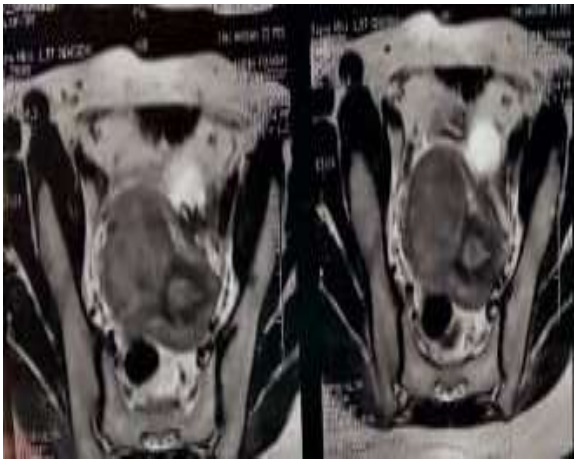
Introduction

According to the 2014 World Health Organisation (WHO) classification, uterine adenofibroma is defined as a benign, biphasic epithelial and mesenchymal, tumour of the uterus [1]. Uterine adenofibroma is an extremely rare neoplasm first described in 1969 by *Laffargue et al.* [2]. This tumour may originate from the endometrium, but it can also (in 10% of cases) develop from the endocervix [3]. In this paper, we report a case of an endometrial adenofibroma with a brief literature review.

Case presentation

We report the case of Mrs F.N, aged 40, nulligest, followed in the oncology department for breast carcinoma of the right breast, for which she benefited from a mastectomy in conjunction with an axillary lymph node removal, then was put on Tamoxifen 5 years ago. As part of her Tamoxifen monitoring, the patient was referred to us for a pelvic ultrasound examination, which revealed a polymyomatous uterus, it was difficult to map the myomas and to explore the endometrium. Note that the patient's clinical examination revealed an enlarged uterus at 12 weeks' amenorrhoea, with a formation reminiscent of a polyp delivered through the cervix being discovered during speculum examination. An MRI scan revealed a polymyomatous uterus, including a 47x54 mm myoma, classified as FIGO 6, in the right anterolateral corporo-fundal region, that was more suggestive of a hypocellular myoma, although an area of sarcomatous degeneration could not be ruled out (Fig. 1). The endometrium was thin on MRI. Our patient was then scheduled for total inter-annexal hysterectomy. Macroscopic examination of the surgical specimen revealed two polypoid lesions in the uterine cavity, measuring 2x1.5 cm and 1x1.3 cm.

Histologically, the tumour had a dual component comprising stretched glandular epithelial structures with no cyto-nuclear atypia, and a moderately cellular stromal contingent with spindle-shaped cells, discreetly atypical, with no evidence of mitosis (Fig. 2). This morphological appearance was compatible with uterine adenofibromas. Furthermore, endometrial samples demonstrated the presence of proliferative endometrium, and the cervical mucosa exhibited no abnormalities. The anatomopathological study of the rest of the hysterectomy specimen revealed the presence of uterine leiomyomas.



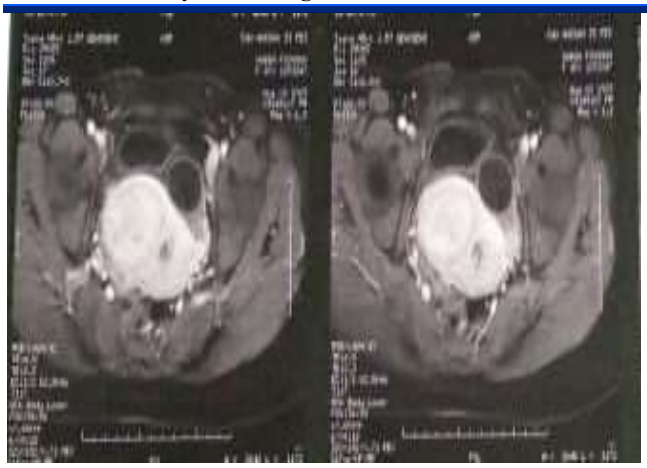
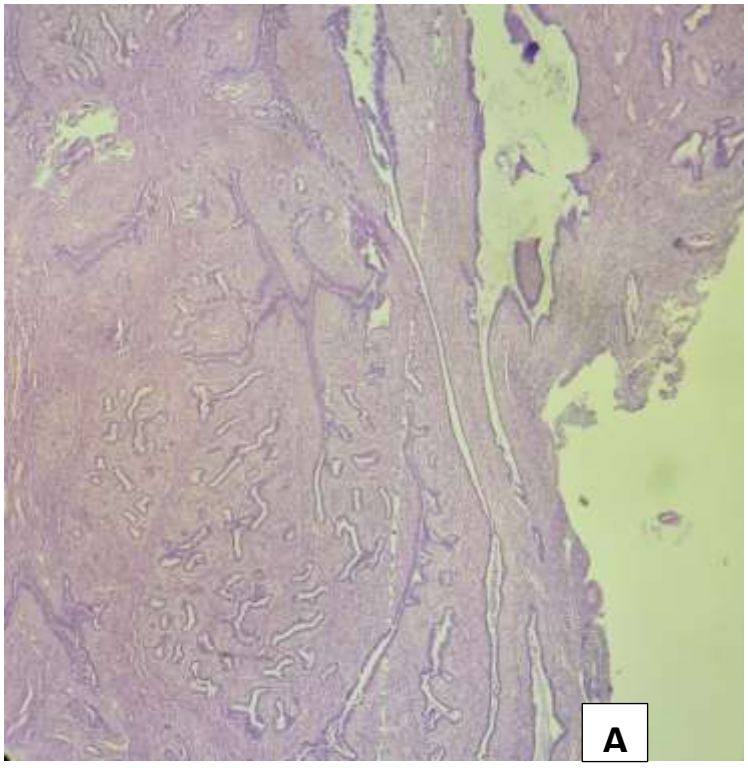


Fig. 1: MRI Scan showing the polymyomatous uterus including a corporo-fundal FIGO 6 myoma suspected of sarcomatous degeneration.

Discussion

Uterine adenofibromas are more likely to occur in post or peri menopausal patients, especially those with a history of polypectomy, with a mean age of onset of 68 years [3]. Yet, our patient is only 40 years old and has no history of polypectomy. Uterine adenofibromas present both clinically and radiologically as benign tumours. Clinically, it is commonly manifested by metrorrhagia, less frequently by pelvic pain or leucorrhoea [4]. In some cases, as in our case, patients may be asymptomatic with the neoplasm being discovered incidentally on radiological examination or in hysterectomy specimens [4]. The physical examination may be entirely unremarkable or, as in the case of our patient, it may occasionally reveal the presence of a polypoid mass delivered through the uterine cervix [4]. On ultrasound, uterine adenofibromas present as a well-limited mass, most often fundal or corporal, with endoluminal projection, containing foci of fluid echo structure [4]. It perfectly simulates an endometrial polyp, a submucosal leiomyoma or, more rarely, a cytogenic chorionic tumour [4]. As clinical and sonographic features are non-suggestive, the pathological examination of a biopsy sample remains the gold standard for the diagnosis of uterine adenofibromas. Macroscopically, uterine adenofibromas appear as tan-brown polypoid tumours, with a maximal diameter varying from 2 to 20 cm and a texture being either soft, firm or elastic [3]. They may contain haemorrhagic foci and cysts responsible for their spongy appearance [3]. Microscopically, uterine adenofibromas are biphasic tumours, composed of a glandular epithelium and a hypocellular stroma [3]. This neoplasm is limited to the endometrium or cervical mucosa, without invasion of the underlying myometrium or cervical stroma [3].



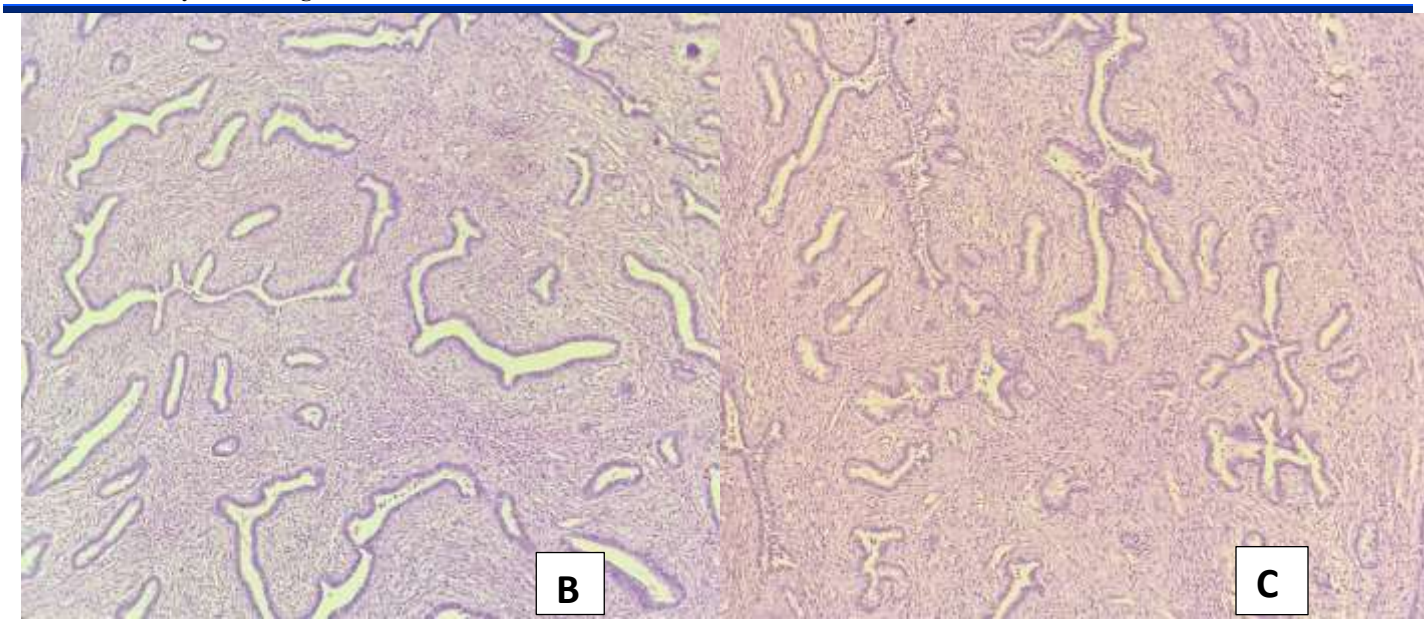


Fig. 2: The microscopic aspect of uterine adenofibroma (HES coloration)

The epithelium is columnar and not notably proliferative, comprising flattened or cuboidal cells [3]. This epithelium is most often of the endometrioid type, although a mixture of various types including endocervical, tubal and squamous, may present in the same tumour [3]. The stroma is typically fibrous, although it can also be cellular, containing collagen and fibroblasts with small and uniform nuclei, without nuclear pleomorphism [3]. From an anatomopathological point of view, adenofibromas must be particularly differentiated from adenosarcomas. The differentiation was based on the greater mitotic activity observed in adenosarcomas, their stromal hypercellularity, the significant nuclear atypia in the stromal cells and the peri glandular cuffing. Nonetheless, in the 2014 WHO classification, there is no mitotic cut off given to make the diagnosis of adenosarcoma, which makes it even more challenging to distinguish from adenofibroma. Therefore, uterine adenofibromas must be considered as a diagnosis by exclusion that can only be diagnosed on complete hysterectomy specimens, rather than on biopsies or polypectomy specimens. Some authors even consider that an adenofibroma is in fact a low-grade or well-differentiated adenosarcoma [5]. Although uterine adenofibroma is a benign tumour, its frequent recurrence [4], its ability to invade and even metastasise [6, 7], together with the possibility of adenocarcinoma developing within the tumour itself or in the surrounding endometrium [4], make total hysterectomy the treatment of choice [4]. Our patient underwent a hysterectomy in view of the initial suspicion of a sarcoma. However, in young patients wishing to conceive, hysteroscopic lumpectomy may be a therapeutic alternative with a long-term follow-up [8].

Conclusion

Uterine adenofibroma is an extremely rare benign tumour. The aim of this work is to remind practitioners, through the report of our case, of the importance of the anatomopathological study being the gold standard in the diagnosis of uterine adenofibromas and to insist on hysterectomy as the treatment of choice for this tumour given its potential for recurrence, invasion and metastasis, as well as the possibility of coexistence with adenosarcoma. However, in young patients wishing to conceive, hysteroscopic lumpectomy may be a therapeutic alternative with a long-term follow-up.

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