

A RARE CASE OF UTERINE INFLAMMATORY MYOFIBROBLASTIC TUMOUR

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OBJECTIVE

We report the finding of a uterine inflammatory myofibroblastic tumor (IMT) in a patient presenting with symptoms commonly associated with leiomyomas. Uterine IMTs are rare, with few cases described in the literature. This case details the presentation, histopathology and management of uterine IMTs, emphasising the need for clinician awareness and thorough histopathological examination for appropriate treatment and follow-up.

CASE REPORT

A 30-year-old nulliparous woman presented with a 15-month history of new-onset dysmenorrhea and prolonged menstrual bleeding without constitutional symptoms.

Transvaginal ultrasound and outpatient hysteroscopy were suggestive of a submucosal leiomyoma arising from the endometrial cavity. A complete transcervical resection of the presumed fibroid was performed with an endometrial biopsy. Histopathology unexpectedly revealed an inflammatory myofibroblastic tumour with a benign, proliferative endometrium.

Uterine IMT is a benign mesenchymal disease with risk of uterine or extrauterine recurrence¹. Postoperative magnetic resonance imaging (MRI) of the abdomen and pelvis along with a computerised tomographic (CT) thorax found no evidence of further local or metastatic disease.

Based on gynaecological oncology multidisciplinary input, the patient will undergo 6-monthly surveillance for 18 months followed by annual surveillance for up to 10 years under the benign general gynaecology team.

Surgical resection or hysterectomy remains the mainstay of treatment with most reported cases achieving remission by 29 months¹. Our patient reported significant improvement in dysmenorrhea and resolution of menorrhagia at 4 months. She remains under active follow-up, awaiting the first surveillance MRI.

DISCUSSION

Uterine IMTs are rare, mesenchymal neoplasms with only 150–200 cases diagnosed annually². They represent a benign condition, though they are locally invasive, and have a potential for distant metastasis or multifocal disease, particularly to the lungs and retroperitoneum³. The majority have been found to arise from the uterine corpus (60%) as found in this case, with reports identifying in association with the placenta during pregnancy (10%), or as a separate mass with wide anatomic distribution (20%)⁴.

Uterine IMTs are estimated to represent 0.1% of leiomyomas⁴. However, unlike benign fibroids, IMTs exhibit variable atypia, gene rearrangements, and a potential for local recurrence or distant metastasis¹. A distinguishing feature is the expression of ALK gene rearrangement⁴.

Surgical resection is the mainstay of treatment, either by hysterectomy or hysteroscopic resection¹. However, systematic therapy can be considered for locally advanced or metastatic cases⁵. Given their unpredictable behaviour, long-term surveillance is necessary. Overall there is a good prognosis for patients with IMTs, with reports of 82.9% five-year event survival and 98.1% overall survival⁵.

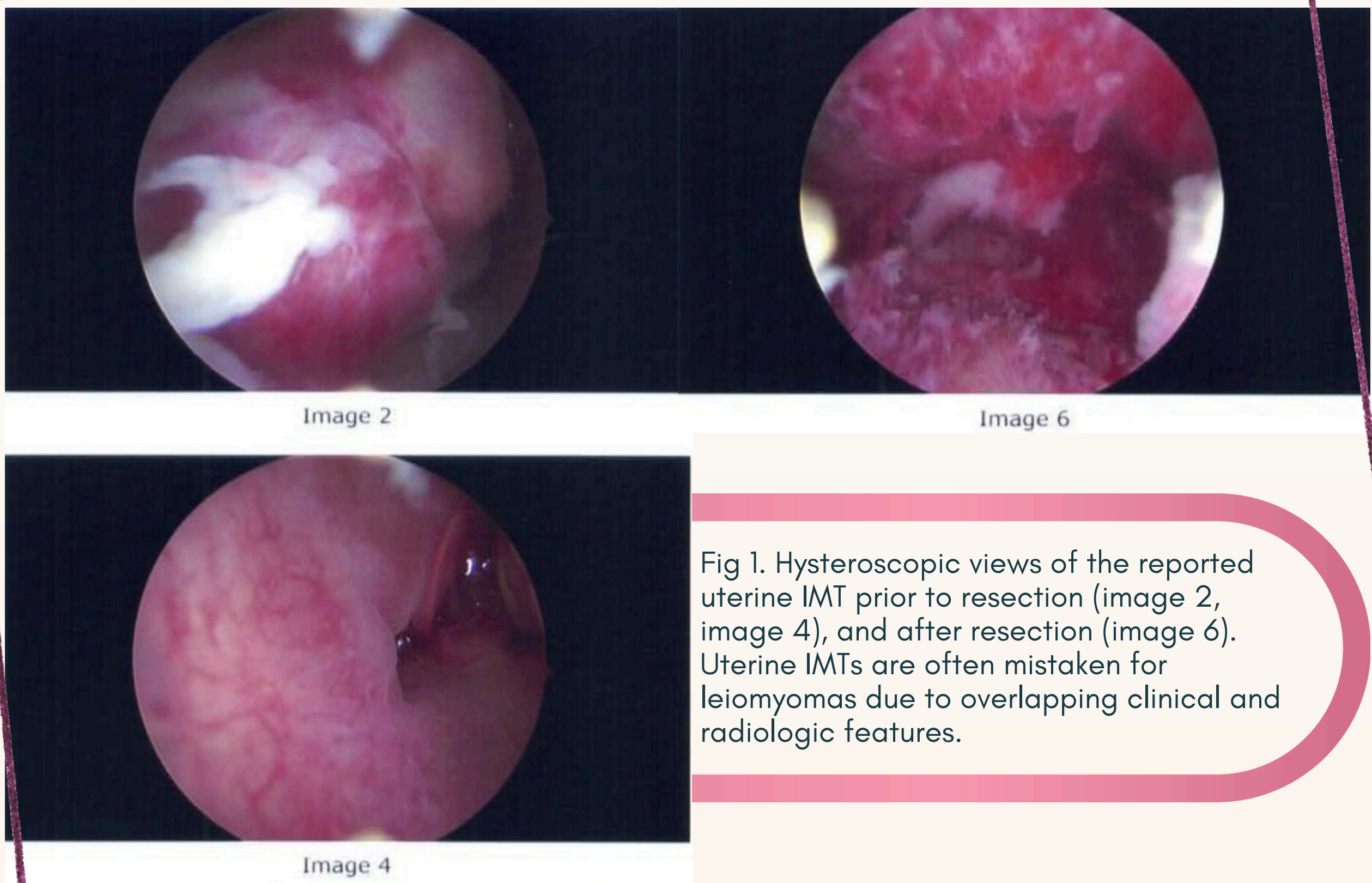


Fig 1. Hysteroscopic views of the reported uterine IMT prior to resection (image 2, image 4), and after resection (image 6). Uterine IMTs are often mistaken for leiomyomas due to overlapping clinical and radiologic features.

IMPACT ON FERTILITY

The patient was nulliparous with a wish to conceive. She was advised to await 18 months of surveillance with no recurrence prior to trying to conceive. Uterine IMTs have been found to be associated with pregnancy, albeit representing smaller, less aggressive tumours⁴. IMTs represent as many as 10% of leiomyomas and 14% of smooth muscle tumours of uncertain malignant potential resected in pregnancy⁴. However, the literature does not describe the impact of pregnancy on recurrence rates nor the impact of IMTs on fertility.

CONCLUSION

Uterine IMTs are rare and can mimic leiomyomas, requiring accurate diagnosis and long-term surveillance. This case highlights the need for histopathological evaluation and standardised follow-up protocols to guide optimal management and future research.

References:
1.Mandato, V.D., Valli, R., Mastrofilippo, V., Bisagni, A., and Aguzzoli, L. (2017) Uterine inflammatory myofibroblastic tumor: more common than expected: Case report and review. [Online] Medicine (Baltimore); 96(48):e8974. doi: 10.1097/MD.0000000000008974. PMID: 29310405
2.Parra-Herran, C., Lee, C.H., Bennett, J.A., and Rabban, J.T. (2020) Inflammatory myofibroblastic tumour. In: WHO Classification of Tumours Female genital tumours. Volume 4, 5th edn. Lyon, France: International Agency for Research on Cancer; pp 298–299.
3.Surabhi, V.R., Chua, S., Patel, R.P., Takahashi, N., Lalwani, N., and Prasad, S.R. (2016) Inflammatory Myofibroblastic Tumours. Current Update. Radiologic Clinics of North America. 54, 553–563.
4.Karpathiou, G., Devouassoux-Shisheboran, M., Stolnicu, S., Chauleur, C., and Péc'h, M. (2023). Uterine inflammatory myofibroblastic tumor. Pathology, Research & Practice.; 242:154335. doi: 10.1016/j.prp.2023.154335. PMID: 36706588.
5.Casanova, M., Brennan, B., Alaggio, R., Kelsey, A., Orbach, D., van Noesel, M. M., Corradini, N., Minard-Colin, V., Zanetti, I., Bisogno, G., Gallego, S., Merks, J. H. M., De Salvo, G. L., and Ferrari, A. (2020). Inflammatory myofibroblastic tumor: The experience of the European pediatric Soft Tissue Sarcoma Study Group (EpSSG). European journal of cancer, 127, 123-129. _