

Ectopic Pelvic Leiomyoma and Diagnostic Difficulty in Imaging: A Case Report from Conakry, Guinea

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Abstract

Introduction: Fibromyomas are benign tumours of frequent uterine location, most often found in black women, whose diagnosis of certainty is histological. The extra uterine location is rare and the physio-pathogenesis is poorly understood. This location poses diagnostic problems on imaging. Management is based on tumour resection. **Observation:** The authors report a clinical case of a 31-year-old woman who was seen for non-febrile hypogastric pain with a large abdominopelvic mass on physical examination. There was no particular history. The biology did not show any abnormality. Ultrasound with Doppler found a mass that was difficult to characterize, and was completed by a CT scan and a pelvic MRI. The management was surgical with an evolution marked by complications. **Conclusion:** Ectopic leiomyoma is a rare condition. This location poses diagnostic problems on imaging. Management depends on the type of extra uterine presentation of the myoma.

Keywords

Ectopic, Histology, Imaging, Leiomyoma, Pelvic

1. Introduction

Myomas are benign tumours, typical of the uterus, found mainly in black women over 30 years of age, with an estimated frequency of 20% - 30% [1]. Their extra-uterine locations are rare and present a greater diagnostic challenge. These histologically benign tumours, which arise from smooth muscle cells, usually occur in

the genitourinary system, but can occur in almost any anatomical site. However, unusual growth patterns may be observed, including metastatic benign leiomyomas, disseminated peritoneal leiomyomatosis, intravenous leiomyomatosis, parasitic leiomyoma and diffuse retroperitoneal leiomyomatosis [2].

The pathophysiology of ectopic leiomyomas remains poorly understood. The main differential diagnoses are fibroma, fibrothecoma and ovarian fibrosarcoma and stromal tumours of the digestive type. The therapeutic management depends on the anatomical setting [3].

We report a clinical case that showed the limitation of imaging, the diagnosis of which was made intraoperatively and confirmed by histology.

2. Observation

Patient H, aged 31 years, was seen for non-febrile hypogastric pain with a satisfactory general condition. On physical examination, a large, hard, painful abdominal-pelvic mass with regular contours was noted.

The woman had four previous vaginal deliveries, the first of which resulted in trauma requiring vaginal reconstruction. The gonadotrophic chorionic hormone level was normal as was the cervical smear. Tumour markers were unremarkable.

Pelvic ultrasound revealed a mixed cystic and tissue mass that was difficult to characterise (**Figure 1**). Abdominal and pelvic CT scan revealed a mixed left latero-uterine pelvic formation with regular contours and heterogeneous enhancement, suggesting an adnexal tumour. This lesion did not show calcification (**Figure 2**). On MRI, a large left-sided abdominal-pelvic mass was noted, hyposignal in T1 and heterogeneous in T2 weighting with no evidence of restriction on diffusion sequences (**Figure 3(a)** and **Figure 3(b)**). This mass slightly compressed the left ovarian vein at the iliac portion with an upstream dilatation. There was no adenomyosis or deep endometriosis. The ovaries were normal. On the basis of this examination, we retained a pedicled fundial myoma with slight remodelling.

After a disciplinary consultation, a surgical management was performed. Exploration of the pelvis after a Pfannenstiel incision revealed an anterior pelvic mass of approximately 14 cm under peritoneal pressure with no obvious connection to the uterus, which was of normal size. This mass was in continuity with the left round ligament of the uterus. We proceeded to excision of the mass followed by peritoneal lavage.

A 400 gramme fresh encapsulated piece was sent for histological study. On section, it is a solid, fasciculated, snow-white lesion with myxoid and necrotic territories. After fixation with formalin, staged samples were taken. Microscopic examination revealed a fasciculated lesion organised in long intersecting bundles of cells, without any atypia. The nuclei are elongated with rounded borders, with fine chromatin and no visible nucleoli. Cytoplasm is abundant and eosinophilic. There is no necrosis or mitosis. However, there are hypo-cellular areas with an

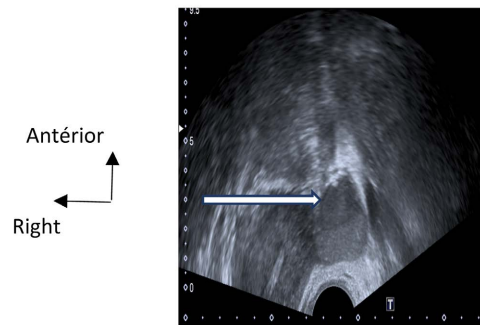


Figure 1. Endocavitary doppler ultrasound showed a heterogeneous left latero-uterine mass (fleche).

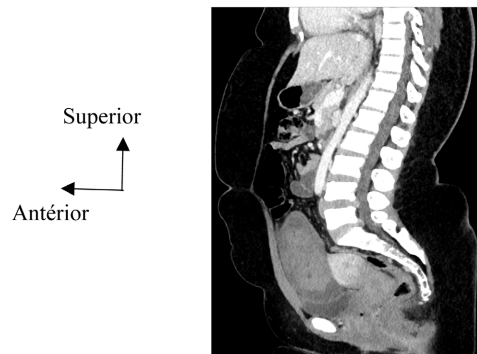


Figure 2. Abdominal and pelvic CT scan with sagittal reconstruction, showing a pelvic mass with heterogeneous enhancement.

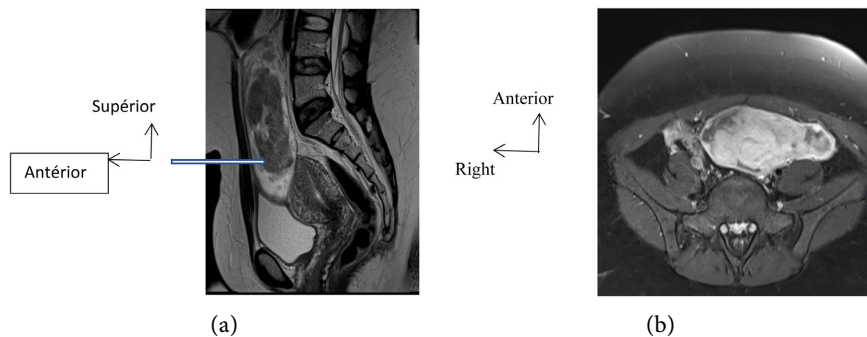


Figure 3. Pelvic T2-weighted MRI, sagittal section (a) and axial FAT SAT with Gadolinium injection (b), showing heterogeneous signal formation and enhancement.

oedematous or myxoid appearance. Immunohistochemical examination showed that the spindle cells were labelled with anti-caldesmon. The lesion was completely removed and the diagnosis was benign remodelled leiomyoma.

The postoperative period was marked by abdominal pain with biological deglobulation requiring a transfusion of 2 red blood cells. An emergency surgical revision was performed after an ultrasound scan at day 2. The intraoperative exploration revealed a large haematoma of the broad ligament with active bleeding which we washed out followed by haemostasis. One week after this last operation, the patient was discharged with favourable results.

3. Discussions

Leiomyomas are solid tumours consisting of fusocellular smooth muscle fibres and collagenous stroma. Most leiomyomas of the genital area arise from the uterus [4]. However, this location is not exclusive, they can develop at the expense of the broad ligament, the round ligament and the ovary, as we found in our observation.

Atypical locations of myomas reported in the literature are benign metastatic leiomyomas, disseminated peritoneal leiomyomatosis, intravenous leiomyomatosis, and retroperitoneal leiomyomas. Roue *et al.* [3] reported three cases of ectopic leiomyomas arising in the round ligament, broad ligament and ovary. Other studies [3] [5] have reported several cases of retroperitoneal leiomyoma (RPL) in the literature between 1941 and 2007.

The pathophysiological origin of isolated leiomyoma is controversial. According to the parasite theory, ectopic leiomyoma is a subserous leiomyoma which, by becoming adherent to adjacent structures, has developed an accessory circulation [6]. For Paal *et al.* [6] this theory has its limitations: it does not explain the existence of ectopic but not pelvic leiomyomas (e.g. retroperitoneum), nor the hormone receptor negative ectopic leiomyomas, or the abdominal or retroperitoneal leiomyomas described in men. The question that arises is whether this development could have been from vascular or extravascular ectopic smooth muscle fibres?

In our observation, the pathogenesis remains difficult as the patient had not developed uterine myomas or operated myomas in her history. The most likely hypothesis in this case would be the development of the myoma from the smooth muscle connective tissue of the ligament or from the smooth muscle fibres of the main ligament artery.

Indeed Kho *et al.* [7] in their series found that 67% of ectopic locations had a history of myomectomy surgery while Poliquin *et al.* found that 40% of retroperitoneal locations had a history of hysterectomy. These findings are different from our clinical case, which could further strengthen the hypothesis of a primary lesion.

The diagnosis of certainty of ectopic leiomyoma is based on histology. Indeed, the circumstances of discovery are very variable [8]: fortuitous, during an investigation of a pelvic mass as in our case, or non-specific symptoms (acute or chronic pelvic pain etc.). Radiological exploration is often limited to give a precise diagnosis. The sonographic appearance is that of a mass with regular contours and variable echostructure [9]. The appearance on CT scan is typical but non-specific with the demonstration of a hypodense lesion, well circumscribed, with heterogeneous hyperdense enhancement [10]. MRI seems to be a promising technique, especially in typical cases, but does not allow a topographical diagnosis [11]. In any case, MRI with reduced sequences is fast and efficient to complement the preliminary ultrasound work-up where the topographic diagnosis was not precise.

Management depends on the type of extrauterine presentation of the myoma.

For isolated leiomyoma, such as this observation, the initial treatment is surgical removal.

4. Conclusion

Ectopic leiomyoma is a rare condition. Imaging plays an important role in their investigation but it seems more difficult to establish criteria of benignity or malignancy. The positive diagnosis of ectopic leiomyoma is based primarily on pathological examination. Management depends on the type of ectopic presentation of the myoma.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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