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Invited Editorial

Atypical manifestations of uterine leiomyomas – expecting the unexpected

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Uterine leiomyomas, or myomas or fibroids, are the most common tumours of the female genital tract, with an estimated incidence of 80% in women of reproductive age [1]. Arising from the monoclonal proliferation of a single myometrial smooth muscle cell, these benign masses vary widely in size, number and clinical presentation [2]. The vast majority of women with fibroids are asymptomatic, while approximately one-third of women experience a spectrum of signs and symptoms dependent on the number, size and uterine location [3].

Fibroids typically present with symptoms such as abnormal uterine bleeding, menorrhagia, pelvic pain and pressure, infertility and early pregnancy complications [1]. However, they can also manifest in an atypical or unexpected manner, which serves as an important reminder that clinicians ought to maintain a high index of suspicion to avoid delays in the diagnosis and treatment of women with this common benign tumour. In this editorial, we highlight the atypical presentations of uterine leiomyomas, their pathophysiological spectrum and their relevance to a multidisciplinary approach to treatment.

Intra-abdominal haemorrhage is an exceedingly rare manifestation of uterine fibroids, with only a few cases reported in the medical literature since 1902 [4]. The aberrant vascular network that feeds subserosal uterine fibroids produces superficial vessels that are weak and prone to rupture [5]. While the specific cause of rupture of these superficial vessels remains unelucidated, trauma, violent coitus, motor vehicle accidents, contact sports, defecation, or pregnancy may provide a reasonable explanation for this rare occurrence [6]. Patients present with sudden-onset severe abdominal pain and profound hypovolaemic shock [4]. In the vast majority of cases, the diagnosis is poorly recognised based on the patient's clinical presentation, and alternative diagnoses such as a ruptured ectopic pregnancy, ruptured ovarian cyst or perforated peptic ulcer are considered [6]. The presence of uterine leiomyoma in women with intra-abdominal haemorrhage of unknown origin may be considered a "red herring" [6]. Resuscitative measures, prompt exploratory surgery with consideration for myomectomy or hysterectomy and a multidisciplinary approach among surgeons, gynaecologists and emergency physicians are imperative to improve patient outcomes [4].

Disseminated peritoneal leiomyomatosis (DPL) is a rare benign disease of unknown aetiology, mimicking disseminated malignancy and affecting women of reproductive age [7]. Leiomyomas are smooth muscle tumours common to the uterus. However, unusual growth patterns may occur, leading to three groups of primary neoplasms: intravenous leiomyomatosis, benign metastasising leiomyomas and disseminated peritoneal leiomyomatosis [8]. Patients with peritoneal leiomyomatosis may present with ascites, peritoneal thickening, and peritoneal or omental nodules, a constellation of signs and symptoms that mimics metastatic ovarian or peritoneal carcinoma [7]. Currently, there are no standardised treatment protocols for DPL, and management consists of an individualised approach that considers the patient's age, symptomatology, and desire for conception [9]. Treatment options include gonadotrophin-releasing hormone (GnRH) agonists, bilateral salpingo-oophorectomy or surgical excision of metastatic nodules [9]. Hysterectomy may be considered if there is accompanying symptomatic uterine leiomyomata [7]. Although DPL is a benign condition, there is a small risk of malignant transformation in 2–5% of cases and long-term follow-up after treatment is recommended [8].

Red degeneration is one of four main types of degeneration that can involve uterine fibroids [10]. It commonly occurs in the mid-second trimester of pregnancy, and is exceptionally rare in non-pregnant women [11]. Red degeneration occurs when an increasing tumour size exceeds its blood supply, leading to haemorrhagic infarction and necrosis of the tumour [10]. Pain is a characteristic sign of degeneration and is usually severe and located at the tumour site [10]. Hysterectomy may be considered for red degeneration of uterine fibroid in non-pregnant women, depending on their desire for future fertility, while conservative management is the standard of care in pregnancy [11].

Another unusual manifestation of uterine fibroids includes compressive mass effects that cause both acute and chronic symptoms [12]. Fibroids have the propensity to attain large sizes that may be

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comparable to a gravid term uterus. The location and size of fibroids may render them immobile in the pelvis, causing them to compress critical structures such as the bowel, ureter, pelvic venous complexes, and even nerves, leading to bowel obstruction, obstructive uropathy and compressive neuropathy [13]. Bowel obstruction is a rare complication of uterine fibroids, with only a few cases documented in the medical literature [14]. Obstructive uropathy occurs as a spectrum of hydroureter, hydronephrosis and acute urinary retention, depending on the location and size of uterine fibroids [15]. Compressive neuropathy from uterine fibroids should be considered in patients who present with acute or subacute paresthesia, muscle weakness or motor impairment, as urgent gynaecological evaluation and surgical management are required to prevent long-term complications [16]. Due to the rarity of this presentation and the need for timely intervention, gynaecologists need to have a high suspicion that a mass of gynaecologic origin can cause varying symptoms due to mass effects.

While uncommon, other potential symptoms of uterine fibroids include a protruding vaginal mass caused by uterine inversion associated with large submucosal polypoidal fibroids [17]. Rarely, large subserosal fibroids may irritate the vagus or phrenic nerve, causing hiccups [13]. Pruritus accompanied by multiple raised skin lesions on the limbs is also unusual and may be the sole symptom of piloleiomyoma [18]. Uterine myomas can coexist with piloleiomyoma [18].

Awareness of both typical and atypical symptoms aids in the early diagnosis of these rare fibroid presentations and allows for appropriate counselling of patients. Imaging tools such as CT and MRI can be helpful when diagnosis proves difficult. There is a risk of compromised function of adjacent organs and higher rates of surgical complications if the fibroid continues to enlarge. Early diagnosis facilitates prompt treatment in the form of medical management, myomectomy when the uterus is still small, thereby preserving fertility or hysterectomy if conception is not desired.

Contributors

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