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A submucosal fibroid's unusual journey to the heart: Intravascular leiomyomatosis with cardiac extension

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Abstract:
Intravascular leiomyomatosis is a rare benign smooth-muscle neoplasm that can extend through the pelvic venous system into the inferior vena cava and right atrium. Hence, a 40-year-old woman with heavy menstrual bleeding, dysmenorrhoea, fatigue and bilateral leg swelling was found to have a submucosal uterine leiomyoma with continuous intravascular extension to the right atrium. Multimodality imaging defined the full extent of disease and enabled combined gynaecological and cardiothoracic surgical planning. Complete resection was achieved histopathology confirmed intravascular leiomyomatosis and no recurrence was identified at 12-month follow-up. Thus, data shows the importance of considering intravascular leiomyomatosis in the differential diagnosis of a right-sided intracardiac mass in women with uterine leiomyoma and of pursuing complete surgical removal when feasible.

Keywords: Intravascular leiomyomatosis, leiomyoma, uterine, inferior vena cava, heart atria, magnetic resonance imaging, hysterectomy

Background:
Intravascular leiomyomatosis is a rare benign smooth-muscle neoplasm characterized by intraluminal growth within venous channels **[1]**. It usually originates in the uterus or pelvic veins and may extend through the inferior vena cava to the right atrium, where clinically important cardiopulmonary complications can occur **[2, 3]**. Premenopausal women and patients with a history of uterine leiomyoma are most frequently affected **[4, 5]**. Although histologically benign, the lesion can behave aggressively because of progressive intravascular extension without conventional tissue invasion **[5]**. Proposed mechanisms include direct extension of a uterine leiomyoma into venous channels and origin from venous-wall smooth muscle, with published evidence more often supporting uterine origin **[6, 7]**. Symptoms depend on disease extent, ranging from gynaecological complaints to venous obstruction and cardiopulmonary manifestations **[8].** Ultrasonography, CT, MRI and echocardiography are important for establishing continuity of the mass and defining its cranial extent before surgery **[9, 10]**. Therefore, it is of interest to evaluate the case of intravascular leiomyomatosis extending continuously from a submucosal uterine leiomyoma through the venous system to the right atrium, with emphasis on imaging, multidisciplinary surgical management and histopathological confirmation.

Case report:
A 40-year-old woman presented with heavy menstrual bleeding and dysmenorrhoea of several months' duration, together with fatigue and bilateral leg swelling. She had no previous history of cardiac disease or thromboembolic events. Examination showed pallor, mild anaemia (haemoglobin 9.8 g per dL) and bilateral pitting oedema; cardiovascular examination was otherwise unremarkable. Pelvic ultrasonography demonstrated an enlarged uterus containing a large heterogeneous submucosal leiomyoma with cystic degeneration **(Figure 1)** and a tubular echogenic structure in the adnexal region raised suspicion of venous extension. MRI showed a uterine mass with signal similar to myometrium on T1-weighted images and mixed high signal on T2-weighted images. A continuous intravascular lesion extended through the uterine veins into the left gonadal vein, renal vein, inferior vena cava and right atrium **(Figure 2).** The mobile intracardiac component remained continuous with the caval component, favouring tumour extension rather than isolated thrombus **[10].** Combined gynaecological and cardiothoracic surgery was undertaken. Hysterectomy was followed by complete extraction of the intravascular tumour through abdominal and thoracic access, yielding a long, firm, whitish venous cast without fragmentation . Histopathology demonstrated interlacing bundles of bland smooth-muscle cells without atypia, confirming intravascular leiomyomatosis **[11] (Figures 3 and 4).** Recovery was uneventful, leg swelling improved and no adjuvant hormonal treatment was given. At 12 months, the patient remained asymptomatic with no imaging evidence of recurrence. The principal differential diagnoses included caval thrombus, right atrial myxoma, tumour thrombus from renal malignancy and intravascular leiomyomatosis.

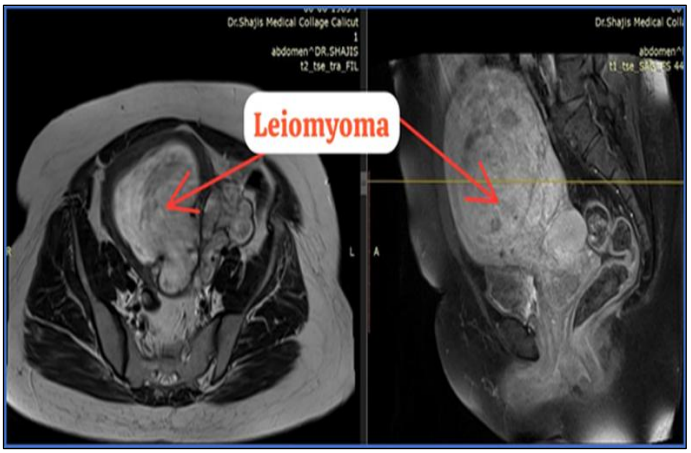


Figure 1: Pelvic imaging showing an enlarged uterus with a submucosal leiomyoma demonstrating cystic degeneration.

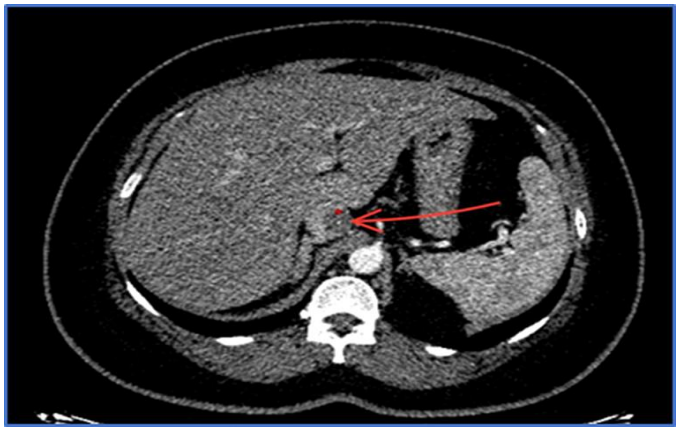


Figure 2: MRI demonstrating continuous intravascular extension of the lesion from the uterus into the gonadal vein and inferior vena cava.

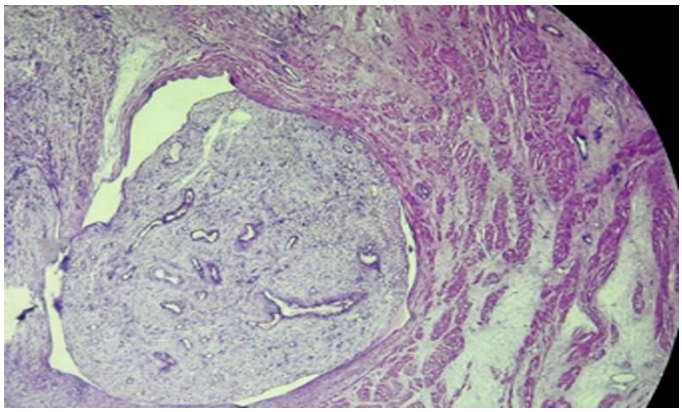


Figure 3: Histopathological section showing intravascular proliferation of bland smooth muscle cells without atypia (hematoxylin and eosin stain, low power).

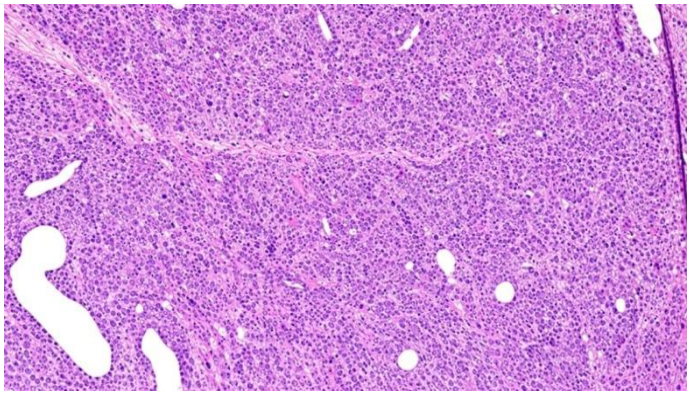


Figure 4: High-power microscopic view showing spindle-shaped smooth muscle cells confirming intravascular leiomyomatosis.

Discussion:

Intravascular leiomyomatosis is a rare but clinically important condition, as extension to the heart can lead to serious and potentially fatal complications **[12]**. Although benign on histology, the lesion can behave aggressively and continue to grow within venous channels **[5]**. Cardiac involvement occurs when the tumor spreads from the uterine or ovarian veins into the inferior vena cava and then reaches the right atrium. Usually, there is no true invasion of the vessel wall, as the mass grows within the lumen as a free intravascular lesion. Upward spread may be related to venous blood flow and negative intrathoracic pressure. Once the heart is involved, patients may present with dyspnea, syncope, or sudden death if untreated **[13]**. Many patients initially present with symptoms of uterine fibroids, most commonly abnormal uterine bleeding **[14]**. Venous spread may cause swelling of the lower limbs, whereas cardiac extension may produce cardiopulmonary symptoms. As these features are nonspecific, diagnosis may be delayed. Imaging is important for diagnosis. Ultrasonography may show abnormal tubular vascular structures. Magnetic resonance imaging is particularly useful for demonstrating the continuity and extent of the lesion from the uterus through the major veins **[15]**. A right atrial mass connected to the inferior vena cava should raise suspicion of intravascular leiomyomatosis rather than thrombus **[16]**. Complete surgical removal remains the mainstay of treatment and extensive disease involving the inferior vena cava or right atrium may require a multidisciplinary surgical approach **[17]**. Multistage or single-stage procedures may be selected according to the extent of cardiac and venous involvement and the patient's surgical risk **[18]**. Recent reports also emphasize careful preoperative imaging and multidisciplinary planning to achieve complete resection in patients with intracardiac extension **[19]**. Published cases of intravascular leiomyomatosis extending through the inferior vena cava into the heart also emphasize imaging of tumour continuity and complete multidisciplinary resection, although the route of venous extension and clinical presentation vary between patients. **[20] [21].** The role of hormonal therapy remains unclear, particularly after complete resection. Long-term follow-up is advised because recurrence has been reported **[16, 17]**. The present case showed the typical pattern of spread from the uterus to the heart. Early diagnosis and complete resection led to a good outcome.

Conclusion:

Intravascular leiomyomatosis may extend from the uterus to the right atrium. Multimodality imaging helps define its continuous venous course. Complete multidisciplinary excision is important when technically feasible.

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