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Intravenous leiomyomatosis as an unusual cause of misdiagnosis of deep vein thrombosis

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Background

Intravenous leiomyomatosis (IVL) is a variant of uterine leiomyoma that permeates into the venous system and grows along the vessel wall. Although histologically benign, biologically it has malignant properties due to intracardiac extension potential, which may lead to heart failure and death. IVL was first described by Birsch-Hirschfeld in 1896 in Germany. Since then about 300 cases of IVL have been reported in medical literature.

Benign metastasizing leiomyoma (BML) is another rare condition which appears as a metastasizing uterine leiomyoma, most often to the lung. Both conditions in one patient are extremely rare and in the literature therefor are about 60 cases reported. [1, 2, 3]. We present the case of a patient with intravenous leiomyomatosis extending to the right heart, metastasizing to the lung and leiomyoma of the left ovary, treated with surgery and hormonal therapy.

Material and methods
A 50-year-old woman, with the history of papillary thyroid cancer treated 7 years earlier, was admitted to a hospital because of heavy menstrual bleeding and anemia. Gynecological ultrasound (US) was performed, which revealed massive uterine fibroids. Hysterectomy was decided. One week after surgery, the patient was re-hospitalized for right-side flank pain and hematuria. Based on computed tomography (CT), hydronephrosis was diagnosed (Fig.1a). Additionally, the CT scan revealed intravenous shadows in the inferior vena cava with canals of proper blood flow, that were diagnosed as deep vein thrombosis (DVT) (Fig.1b). The additional diagnostic procedures for the chest and pelvic area were implemented. The examinations showed venous masses extending from the right atrium of the heart to the common iliac veins (Fig.1c,d).

Although the patient did not show any clinical symptoms of DVT and D-dimers levels were low - 302 ug/l, anticoagulation therapy was implemented. Moreover, chest-CT revealed few nodules measuring up to 13 mm in the right and left lung described as metastases of unknown origin (Fig.1e).

PET-CT did not show any lesions with higher 18-F-FDG uptake. After one month of pharmacological therapy, no regression of intravenous masses was observed. Surgical removal of the right atrial and iliac vein masses was executed. During sternotomy and laparotomy the myomatous tissue was removed from the heart, iliac veins and IVC. Histopathology confirmed the diagnosis of intravenous leiomyomatosis. Two months after surgery Doppler ultrasound examination revealed new tumor tissue growing in IVC (Fig.1f); one year after surgery IVL of inferior vena cava and iliac veins recurred. Fortunately, the heart chambers were still clear. Moreover, contrast-enhanced MRI and ultrasound examinations showed new zones of similar morphology in the left ovary (Fig.1g, h). Currently, the patient receives anti-estrogen therapy, as there are studies showing a hormonal undertow of the disease; [4] she wheezes slightly yet shows no clinical features, and remains under observation.
Concluding, although IVL is a very rare entity, it could be underestimated due to its possible oligosymptomatic course. [5] IVL should always be considered in middle-aged women with the history of uterine fibroids and characteristic intravenous masses, to avoid misdiagnosis (deep vein thrombosis or metastases) and overtreatment.

References:


Figure 1 (a). – Contrast enhanced computed tomography. Hydronephrosis of the right kidney (arrow) visible on axial image obtained in aortic phase. (b). – Contrast enhanced computed tomography. Persistent filling-defects within the inferior vena cava (arrow). Axial image in porto-venal phase. (c,d). – Contrast enhanced computed tomography. Venous masses (arrows) extending from the right atrium of the heart to the common iliac veins on axial (d) and coronal (c) images in porto-venal phase. (e).- Chest computed tomography scan. Bilateral lung nodules were visualized (arrows), lung window. (f).- Color Doppler sagital image. Return of leiomyomatosis within inferior vena cava (arrows). (g). – Contrast enhanced magnetic resonance image. Leiomyoma tissue (arrows) enhances after contrast administration in the left ovary. Axial image. (h) - Color Doppler axial image. Left ovarian leiomyoma masses (arrows).