

Aseptic cerebral venous thrombosis associated with a gastric leiomyoma

Maxence Lefebvre¹, Patrick Yengue¹, Jean-François Gallez¹, Patrice Borgies²

(1) Department of Gastroenterology, (2) Department of Internal Medicine, Centre Hospitalier de Wallonie Picarde, Tournai, Belgium.

To the Editor,

In January 2015, a 38-year-old woman consulted a neurologist for heavy diffuse headache, visual field defects, transient left paresthesia and seizures. She had no fever nor other complaint. She used DIV NUVARIN, an intravaginal ring, as contraceptive method. Her past medical history was only marked by an appendectomy during childhood. There was no objective neurological abnormality and the remainder of the examination was normal. CRP was slightly increased (21 mg/l) and the WBC count was 11680/mm³ with 92% neutrophils. Cerebral magnetic resonance revealed a superior sagittal sinus thrombosis without any abnormality inside the brain (Fig. 1).

Standard tests of coagulation were normal. Tests for proteins S, C and antithrombin deficiency, antiphospholipid antibodies and homocysteinemia were negative. There was no factor V Leiden mutation, no biological nor clinical evidence for acquired prothrombotic condition.

The patient was started on therapeutic anticoagulation with acenocoumarol and her clinical condition quickly improved.

To exclude a cancer-associated thrombosis a FDG-PET-CT-scan was performed. This revealed a large

tumor next to the anterior wall of the stomach with low avidity for FDG (maximum standard uptake value (SUVmax): 3.9) and diffuse uptake of the right colon (SUVmax: 6.5) (in comparison, the SUVmax of the liver was 3.6). Gastroduodenoscopy was normal. Colonoscopy was normal and biopsies revealed no microscopic colitis. Contrast-enhanced computed tomography demonstrated an exophytic 6 cm-large extragastric tumor (Fig.2). On endoscopic ultrasonography (EUS), the tumor was hypoechoic, homogenous, with some small cystic lesions, and arised from the fourth hypoechoic wall layer, without adjacent lymph nodes nor ascites. EUS-guided fine needle aspiration (22 G) showed spindle

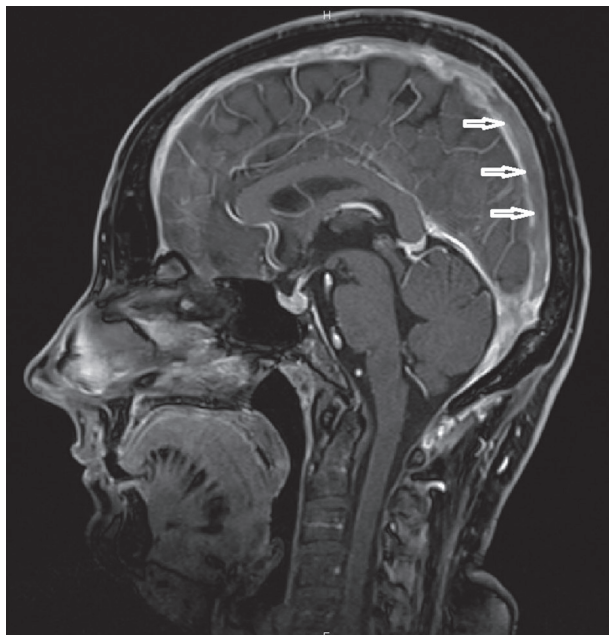


Fig. 1. — Cerebral magnetic resonance revealed superior sagittal sinus thrombosis (white arrows).



Fig. 2. — Computed tomography demonstrated a 6 cm-large extrinsic gastric tumor.

Correspondence to : Dr M. Lefebvre, Rue de l'Escalette, 56 7500 Tournai. Fax: 069/258614
Email : lefebvremaxence@hotmail.com

Submission date : 25/02/2017
Acceptance date : 22/05/2017

cells with cigar-shaped nuclei. A laparoscopic wedge resection was performed. Immunohistochemical stains were diffusely positive for smooth muscle actine and desmin and negative for CD117 and DOG1, with a low mitotic rate Ki-67 (1%), confirming a benign leiomyoma.

Two years later the patient was asymptomatic. Anticoagulant therapy was stopped.

Discussion

Cerebral venous thrombosis (CVT) is an uncommon condition (incidence of 0.5 to 1.5 per 100000 population per year). Etiologies include local causes (central nervous system infection, head trauma or surgery) and classical cause of venous thrombosis (1). In this case, it is clear that the use of a vaginal contraceptive device increased the thrombotic potential without reaching the thrombogenic threshold if no other pro-thrombotic condition is present. It was essential to carry out a complete haemostasis assessment and search an underlying inflammatory or tumoral illness. Leiomyomas are benign smooth muscle tumors. The US-endoscopic aspect is a homogeneous hypoechoic lesion, developed from the second or the fourth layer (muscular mucosal or muscular) (2). On immunohistochemistry leiomyoma's cells are strongly positive for desmine and actine expression and negative for CD117 and DOG1 conversely for GIST (3). The

absence of nuclear atypia as well as the absence of important mitotic activity distinguishes between leiomyoma and leiomyosarcoma. A cause-effect link between gastric leiomyoma and CVT is difficult to establish. Just one case of paraneoplastic thrombosis associated with gastric GIST has already been described (4) as well as one case of paraneoplastic syndrome associated with ovarian leiomyoma (5). To our knowledge, the association between gastric leiomyoma and CVT has not yet been reported.

References

1. COUTINHO J.M., FERRO J.M., CANHÃO P., BARINGARREMENTERIA F., CANTU C., BOUSSER M.G. *et al.* Cerebral venous and sinus thrombosis in women. *Stroke*, 2009, **40** : 2356-61.
2. PAPANIKOLAOU I., TRIANTAFYLLOU K., KOURIKOU A., RÖSCH T. Endoscopic ultrasonography for gastric submucosal lesions. *World J. Gastrointest. Endosc.*, 2011, **3** (5) : 86-94.
3. GONZÁLEZ-CÁMPORA R., DELGADO M.D., AMATE A.H., GALLARDO S.P., LEON M.S., BELTRÁN A.L. Old and new immunohistochemical markers for the diagnosis of gastrointestinal stromal tumors. *Anal. Quant. Cytol. Histol.*, 2011, **33** : 1-11.
4. DES GUETS G., CUENCA X., MORERE J.F. Thrombophlébite cérébrale associée à un GIST. *Sang Thrombose Vaisseaux*, 2007, **19** (7) : 389-91.
5. YUMRU A.E., BOZKURT M., AYANOĞLU Y.T., AYHAN I., INCI COSKUN E., BATTAL HAVANE S. The relation between the presence of a giant primary ovarian leiomyoma and the occurrence of epilepsy as a paraneoplastic syndrome. *Arch. Gynecol. Obstet.*, 2010, **281** (3) : 531-4.