

IMAGEM EM NEUROLOGIA/IMAGE IN NEUROLOGY

Infectious Meningovascularitis Secondary to a Rectothecal Fistula in a Patient with Crohn's Disease

Meningovascularite Infeciosa Secundária a uma Fístula Reto-Tecal num Doente com Doença de Crohn

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We present a case of a 52-year-old male, with a diagnosis of Crohn's disease with multiple fistulae, with a previous pelvic magnetic resonance imaging (MRI) revealing several bilateral perianal fistulae extending to the pre-sacral region, and numerous collections in the retrosacral region, compatible with suppurative hidradenitis. He had never been treated with any immunosuppressive treatment nor had been submitted to surgical intervention.

He was admitted to the emergency room with a subacute onset of obtundation, fever (38.9°C), and right hemiparesis with hypoaesthesia. Here, he had a first tonic-clonic seizure of unknown onset. Blood tests revealed leucocytosis with neutrophilia, and an increased C-reactive protein (179.3 mg/L, normal range 0-5 mg/L). Spinal computed tomography (CT) scan revealed the presence of several scattered subarachnoid aerocoles. Furthermore, at lumbar-sacral level a rectothecal

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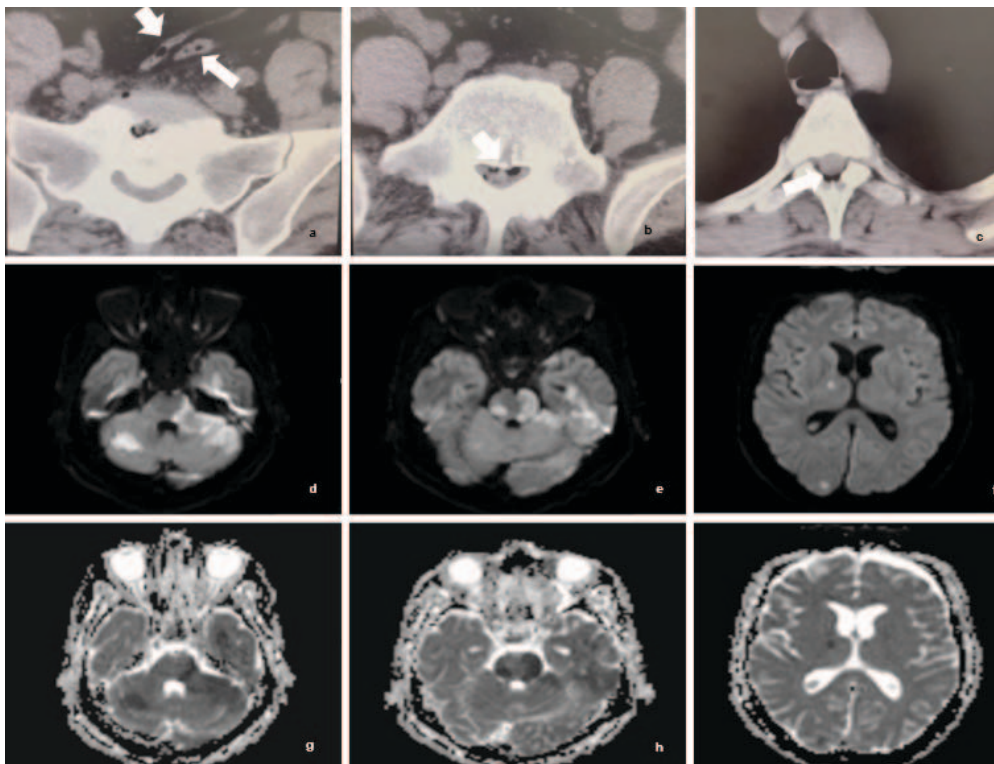


Figure 1. Lumbar spinal CT shows a rectothecal fistula (a - white arrows), and air inside the spinal canal (b and c - white arrows). Head MRI with several diffusion restricted (d-f) and ADC hypointense lesions (g-i), consistent with ischaemic lesions in the setting of an infectious vasculitis.

fistula could also be seen (**Fig.1 A-C**).

Over the next hours, he became stuporous and progressed to coma, with the need for mechanical ventilation. Levetiracetam and empirical antibiotic therapy (ceftriaxone, vancomycin and metronidazole) were started.

The patient was then admitted in an Intensive Care Unit (ICU). He was comatose (Glasgow Coma Scale 3T), with non-reactive mydriatic pupils, absent oculocephalic and corneal reflexes. He was also hemodynamically unstable, requiring vasopressor support. Lumbar puncture was performed, and a thick greenish fluid was collected, making cytochemical analysis impossible. Cerebrospinal fluid (CSF) culture revealed the presence of *Streptococcus constellatus* and *Bacteroides fragilis*, both belonging to the colonic flora. Blood cultures were unremarkable.

Brain MRI revealed multiple intra-axial diffusion restricted lesions along the cerebellum, pons, midbrain and both cerebral hemispheres, with no clear T2/FLAIR hyperintensity, compatible with acute ischaemic lesions in several vascular territories (**Fig.1 D-I**). There was no evidence of large vessel occlusion, microbleeds or meningeal enhancement.

Clinical condition progressed to multiple organ failure and death was declared three days after ICU admission. An infectious meningovasculitis secondary to a rectothecal fistula was considered the most likely cause of death.

Vasculitis due to central nervous system (CNS) infections is a well-recognized complication of a myriad of microorganisms, being most associated with bacterial (*Streptococcus pneumoniae*, *Neisseria meningitidis*, *Mycobacterium tuberculosis*, spirochetes), viral (herpes family viruses, retroviruses, hepatitis agents) and fungal agents, usually reaching the CNS through haematogenous or contiguous spread.¹ This case represents a rare example of a polymicrobial meningoencephalitis secondary to a rectothecal fistula, supported by the identification on CSF cultural examination of two bacteria commonly present in the colonic microbiota. The sterile blood cultures make a haematogenous spreading a less likely

mechanism of infection.

To our knowledge, there are only a few similar cases ever been published, mainly related with colorectal surgery complications^{2,3} or neural tube malformations.⁴ The identification of this phenotype in patients with Crohn's disease is of utmost importance since appropriate medical and surgical treatment of these fistulae can prevent this rare, but ominous, neurological complication. ■

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