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Review

Endometriotic lesions of the lower troncular nerves

Endométriose et atteinte tronculaire des nerfs du membre inférieur

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ABSTRACT

Although exceptional, endometriotic lesions of the troncular nerves of the lower limb may occur and are often diagnosed with delay. We report, hereby, the first case of femoral nerve endometriosis the treatment of which consisted of radical resection with femoral nerve transplant. We completed a review of the literature on sciatic nerve endometriotic lesions and discussed the physiopathology and surgical treatment.

RÉSUMÉ

Bien qu'exceptionnelles, les atteintes endométriosiques des nerfs tronculaires des membres inférieures sont possibles et souvent de diagnostic tardif. C'est ainsi que nous rapportons le premier cas d'endométriose du nerf fémoral et son traitement chirurgical avec greffe de nerf fémoral. Après une revue de la littérature des lésions du nerf sciatique, nous en discutons la physiopathologie et le traitement chirurgical.

1. Introduction

Endometriosis is associated with the presence of ectopic endometrial tissue for approximately 10% of women of child-bearing age. The most common symptoms of this disease are chronic disabling pelvic pain, severe dysmenorrhea, dyspareunia, colorectal disorders and infertility. It has been known for years that there are neurotropic endometriotic lesions [1]. We report here the first case of infiltration of the femoral nerve by an endometriosis nodule and then we review the different cases of infiltration of the lower limbs nerve trunks published in the literature.

2. Case report

A 29-year-old patient with no relevant past medical history but with primary infertility complained of chronic pelvic pain with dysmenorrhea, severe dyspareunia associated with catamenial episodes of right cruralgia, and partial sensory and motor loss in the femoral nerve territory.

A CT scan examination revealed a proximal lumbar psoas muscle hypertrophy. Because of the perseverance of the symptomatology and the worsening sensory-motor deficit, further exploration was performed and an MRI was prescribed.

The MRI revealed a severe pelvic endometriosis, a right uterosacral ligament attached to the anterior rectum, an endometrioma on the left ovary, and many adnexal adhesions. In the iliopsoas muscle region, a heterogeneous fibrous nodule of 29×42 mm was found between its two bellies containing a hemorrhagic locus that led us to suspect an endometriotic origin (Fig. 1).

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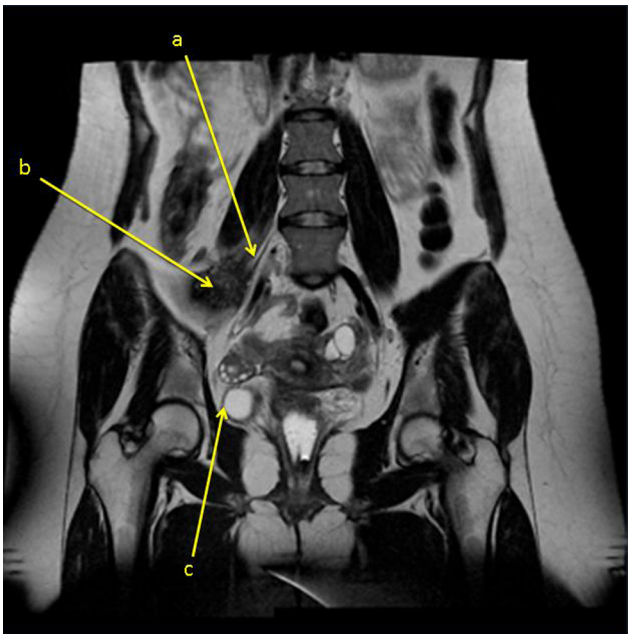

Fig. 1. Coronal pelvic MRI. a: femoral nerve; b: endometriotic lesion; c: right endometrioma.

The initial treatment consisted of a surgical resection of the pelvic endometriosis and a biopsy of the psoas lesion by laparoscopy. A treatment by GnRHa was started two months prior to surgery. Laparoscopic exploration discovered severe pelvic endometriotic lesions of stage 4 rAFS, an inflamed and adherent peritoneum in the paracolic gutter in the region next to the iliopsoas endometriotic lesion. An inflamed appendix was found in a retrocaecal position. An appendectomy was performed followed by an iliopsoas nodule biopsy. The histology confirmed the endometriotic nature of the lesion, the massive infiltration of the femoral nerve and the infiltration of the appendix. Immediately following the operation, the patient showed an aggravation of the

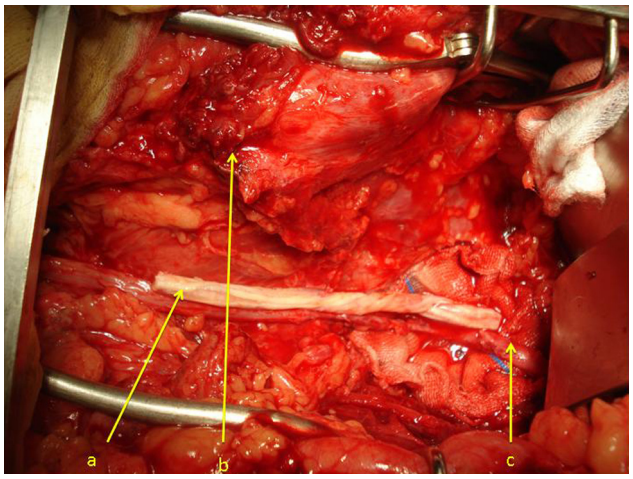

Fig. 2. Lateral surgical view. a: sural transplant; b: psoas muscle; c: femoral nerve.

partial sensory-motor deficit in the right femoral nerve territory. An injected CT scan discovered a psoas muscle haematoma, which explained the symptomatology. The neurological examination found an amyotrophy of the right quadriceps, an absent right knee reflex, and hypoesthesia of the medial side of the anterior thigh and leg. The electromyogram discovered a complete sensory and motor deficiency of the right femoral nerve (Fig. 2). A complete resection of the nodule and a femoral nerve transplant by laparotomic route was proposed to the patient as a treatment option. The patient agreed to it and a gynaecological and neurosurgical team performed the complete resection of the nodule together with part of the femoral nerve. Two sural and musculocutaneous nerves were used to reconstruct the femoral nerve. No complication was observed postoperatively and a specific rehabilitation program was started. GnRHa treatment was maintained to avoid any cyclical micro-haemorrhage and diminish risk of recurrence. One year later, the patient showed signs of partial recovery and was walking without crutches. The GnRHa treatment was interrupted

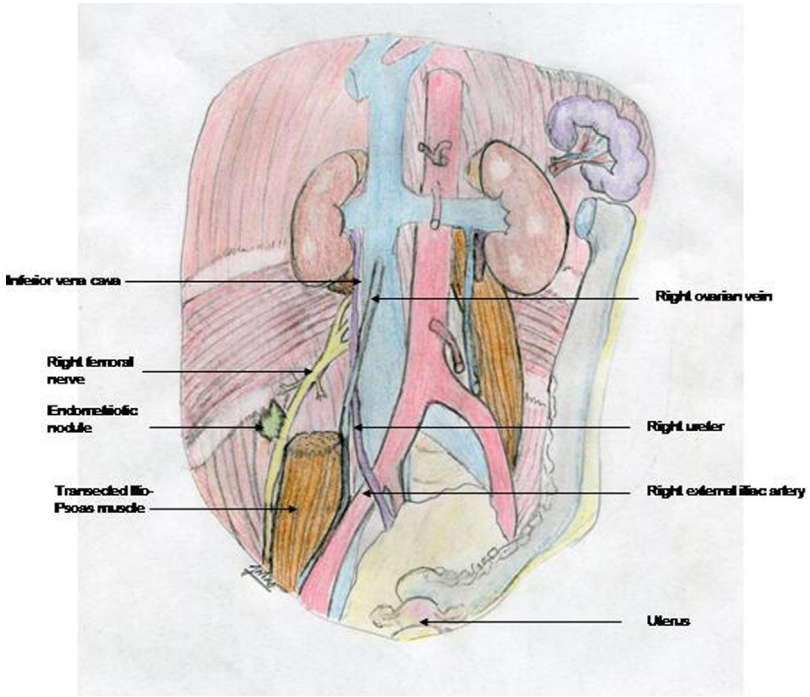

Fig. 3. Frontal view of the femoral nerve. Yellow: femoral nerve.

and a spontaneous pregnancy occurred 4 months later. She vaginally delivered a healthy baby weighting 4040 grams without any complications.

This is the first case of a femoral nerve endometriosis described in the literature.

The femoral nerve remains extra peritoneal all along its trajectory towards the lower limb (Fig. 3). It originates from second, third and fourth lumbar roots. It crosses posterior to the psoas major directed laterally before continuing on the antero-medial surface of the iliacus. It passes under the inguinal ligament, lateral to the femoral vessels and the iliopectineal arch. It then pierces the iliopsoas sheath formed by the two bellies of the iliopsoas muscle. The nerve then terminates by dividing into four branches, the quadriceps nerve, the medial musculocutaneous nerve, the lateral musculocutaneous nerve and the saphenous nerve.

3. Sciatic nerve endometriosis

Endometriosis in contact with the sciatic nerve is a rare site of the disease. A literature review in 2003 found 63 cases in the English literature [2]. Sciatic nerve involvement in endometriosis in this series was predominantly on the right (67.2%), although Possover et al. [3] found 88% of cases to be on the left in a series of 27 patients with selective sciatic nerve disease. The symptoms of isolated sciatic nerve endometriosis are catamenial L5-S1 sciatica, gluteal pain and occasionally loco-motor deficits (from foot) but never include urinary disturbance (due to lower involvement of S2-S3) [4]. Without appropriate management it may progress to loss of sensation and motility in the territory concerned [5]. The catamenial sciatica may be associated with signs of other nerve trunk involvement (pudendal, inferior hypogastric plexus [4]) and other suggestive symptoms of endometriosis (dysmemorrhea, dyspareunia and infertility). Over time the sciatic pain may become permanent, often with a preserved or increased catamenial feature. The diagnosis of endometriosis in contact with the sciatic nerve should, therefore, be considered in a young woman with catamenial sciatica, particularly if usual investigations find no cause for the sciatica.

4. Discussion

4.1. Pathophysiological hypotheses for nerve trunk involvement

The entirely retroperitoneal path of these nerves raises the question of the pathogenesis of the endometriosis, which is still not well explained. Sampson's initial hypothesis by which it was due to endometrial reflux through the fallopian tubes during menstrual cycles may explain the two most common forms of endometriosis, endometriomas and peritoneal endometriosis but cannot easily explain sub-peritoneal endometriosis and particularly the case of our own patient. The other theories of coelomic metaplasia and haematogenous spread are difficult to consider [6–8].

In nerve involvement from endometriosis, particularly the sciatic nerve described on several occasions especially by Possover, correlations have been established with pelvic disease, for example, with the pocket sign described for the first time in 1962 [9] and due to peritoneal invagination, which is often located on the posterior part of the ovarian fossa. Sites of endometriosis are often found at the base of these peritoneal pouches [10]. This theory may possibly apply to our patient taking account of the appendicular site of the endometriosis lying behind the caecum.

Signorile et al. [11] further support the theory that deep endometriosis is due to defective embryogenesis and migration of endometrial tissue and found ectopic endometrial tissue recognized on immunohistochemistry along the path of the malarian ducts in 10% of human foetuses studied. This theory may explain the exclusively retroperitoneal involvement.

4.2. Further investigations of nerve trunk disease

Electrophysiological studies (distal motor and sensory peripheral nerve latent period, sacral latent periods, EMG, cortical evoked potentials and cutaneous sympathetic potentials) provide a positive diagnosis of peripheral nerve damage and locates the disease to the nerve trunk or nerve root and is useful in establishing the prognosis for recovery. These investigations do not indicate the mechanism of the damage [12,13].

If performed by a radiologist experienced in the diagnosis of endometriosis and guided by clinical findings, MR can demonstrate endometriosis lesions along nerve pathways. It can also be used for the full assessment of other concomitant endometriosis lesions.

Several treatments may be tried in order to offer patients relief. Physiotherapy with stretching exercises and osteopathy are often effective although there are few studies on these therapies [14]. Medical treatments involve WHO steps 1 to 3 analgesics which are often relatively ineffective on neuropathic pain but should be tried. Tricyclic antidepressants are effective on neuropathic pain. Doses are adjusted in progressive steps until the desired effect is obtained. They should not be combined with Tramadol. Antiepiletics are effective on the shooting component of the pain and LHRH analogues are a therapeutic test supporting that the damage is due to endometriosis.

4.3. Surgery for nerve trunks

In isolated sciatic nerve endometriosis, Possover [3] found in his series that the lesions appeared to develop in the sheath of the nerve itself and then caudally through the greater sciatic foramen. He, therefore, recommended that a transgluteal approach be avoided as this risks not being complete and opted rather for a retroperitoneal approach allowing intrafascicular neurolysis of the antero-posterior sciatic nerve with resection of the damaged or invaded parts of the nerve. This resection technique is unlike the surgery for endometriosis in the sacral plexuses, which do not require dissection and decompression. The retroperitoneal approach to the sciatic nerve involve dissecting the interiliac space, detaching the pelvic wall adipose-lymphatic tissue and then following the posterior part of the obturator internus muscle towards the sciatic spine.

For isolated femoral nerve involvement, we recommend that the peritoneal sack be pushed back medially in order to release the iliopsoas muscle, which is divided and then raised in order to release the femoral nerve.

5. Conclusion

Lower limb nerve trunk endometriosis is a rare finding in the literature. We describe here the first case of deep endometriosis affecting the femoral nerve. The incidence of this disease must, however, be underestimated in light of the number of patients who complain of this type of chronic pain. The leading problem is the diagnosis as the neurological symptoms are catamenial. Management is multidisciplinary between gynaecological surgeons, neurosurgeons and radiologists and high quality MR is important to precisely identify the site of the lesion. The second difficulty, of course, is the surgical approach which may require nerve grafting and therefore long term follow-up to confirm that nerve functions have fully recovered.

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

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