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CASE REPORT

The Diagnostic Dilemma of a Genitofemoral-ilioinguinal Overlap Syndrome

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ABSTRACT

Background: It is sometimes difficult to clinically delineate genitofemoral and ilioinguinal neuralgias because of the overlap in the clinical symptomatology.

Case description: A young male with a past history of transurethral removal of ureteral calculi presented with severe, debilitating neuropathic pain in the groin. He had hyperpathia and allodynia in the distribution of genitofemoral nerve, and hence a clinical diagnosis of genitofemoral neuralgia was made. An ultrasound guided diagnostic block of the genitofemoral nerve with local anesthetic produced only a mild reduction in pain (VAS 2 reduction). A repeat diagnostic block of the ilioinguinal nerve produced complete resolution of pain.

Literature search showed a limited number of case reports of ultrasound guided blocks for genitofemoral neuralgia; and overlap syndromes have been addressed with differential nerve blocks.

Clinical relevance: Our hypothesis is that an aberrant reinnervation from the ilioinguinal to genitofemoral nerve may present with features favoring a genitofemoral neuralgia. In situations where radiofrequency treatment is being considered, such cases may pose a failed intervention if both nerves are not separately targeted.

Keywords: Genitofemoral, Ilioinguinal, Neuralgia.

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INTRODUCTION

Neuropathic pain is variously described as a stabbing, burning, shooting or pricking sensation. In contrast, non-neuropathic pain is a constant dullache over the entire groin area with no specific trigger point and is usually aggravated by strenuous exercise. Patients commonly describe it as a gnawing, tender, pulling or pounding sensation.[1] Neuropathic pain of ilioinguinal neuralgia is usually distributed along the sensory innervations of the affected nerve(s) and can be reproduced by tapping the skin medial to the anterosuperior iliac spine or over an area of local tenderness (Tinel's test). The clinical differentiation of ilioinguinal, iliohypogastric and genitofemoral neuralgia is difficult, frequently resulting in misdiagnosis and inappropriate treatment.[2,3]

This is because of the overlapping sensory innervations of these three nerves, peripheral communication between their nerve twigs and, most importantly, their common roots of origin (Fig. 1).[4] Genitofemoral neuralgia results in a chronic neuropathic pain along the distribution of the genitofemoral nerve. The nerve has a femoral branch and a genital branch, each with an area of supply which includes the upper part of the thigh, the mons pubis and scrotum in male and labia in female. Genitofemoral neuropathy has been described following inguinal and femoral region surgeries, such as herniorraphy.[5]

The purpose of this report is to highlight the diagnostic dilemma which, unless addressed, may result in failure of therapeutic interventions targeting a single nerve in patients presenting with overlap syndromes.

CASE REPORT

A young male presented with a one year history of severe, jabbing pain in the right inguinal, scrotal and upper thigh region. The visual analog scale (VAS) was 9/10. The pain is diffuse and is aggravated on movements and on touching the area of pain. There was no radiation of pain or backache. There was no dysuria, hematuria, local redness or swelling. There was a past history of a transurethral removal of ureteral calculi. He was given antidepressant and anticonvulsant medications in optimal doses, which did not produce any significant benefit. General physical examination was normal. Neurological examination revealed a hyperpathia and allodynia at the middle of the inguinal ligament, scrotum and medial thigh. Other systems were normal.

He was admitted at another center and a genitofemoral nerve block was given, which produced only a mild

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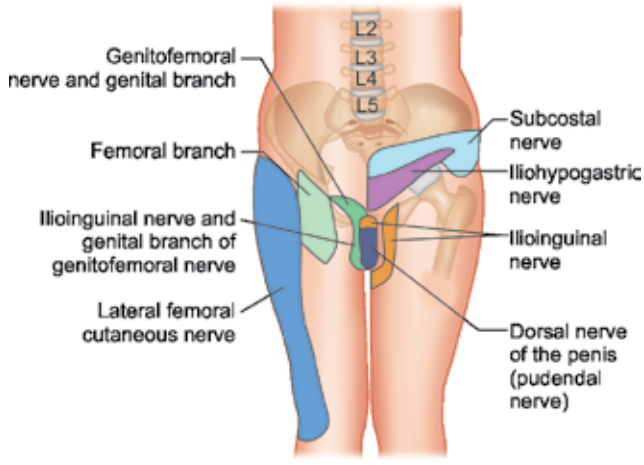

Fig. 1: Territory of supply of ilioinguinal and genitofemoral nerves

reduction (VAS 2) in pain. This history, along with the distribution beyond the distribution of the genitofemoral nerve, was suggestive of a possible overlap in the genitofemoral-ilioinguinal nerve territories.

Blood examinations, magnetic resonance imaging (MRI) of the spine and pelvis and nerve conduction study were normal. Paraneoplastic work-up including computed tomography (CT) abdomen and pelvis were negative. After admission to this hospital, he underwent a USG-guided block of the genitofemoral nerve with lignocaine, to which he reported mild relief (Figs 2 and 3). A second USG-guided block with lignocaine was given, this time targeting the ilioinguinal nerve. There was a complete resolution of pain following both the blocks.

DISCUSSION

This case adds on to the presumption that a dual neuropathy at overlapping territories may be difficult to manage by invasive techniques, unless they are addressed separately. This is because of the occasional communication between both nerves.

Genitofemoral and ilioinguinal neuralgias can occur after inguinal herniorrhaphy, appendectomy, cesarean section, trauma to lower quadrant of the abdomen or inguinal regions. The etiological differential diagnosis of ilioinguinal neuralgia includes nerve damage at the first lumbar root and hip disease.

In those cases with ilioinguinal neuralgia, a nerve block is carried out at a point 2 or 3 cm medial and inferior to the anterior superior iliac spine, and cases with genitofemoral neuralgia, either a block is done on the inguinal ligament or a paravertebral block of L1 and L2 can also be performed. If ilioinguinal nerve entrapment, injury or a neuroma in the abdominal wall is responsible for the pain, a local block of that nerve through the lower anterior abdominal wall should alleviate the symptoms. If the pain is not relieved, genitofemoral injury could be responsible for the pain.[6,7]

There are various treatment options which include pharmacologic treatment, interventional techniques (selective nerve blocks), minimally invasive procedures (pulsed radiofrequency or peripheral neurostimulation), and surgical intervention in genitofemoral and ilioinguinal neuralgia.[8-10] A number of drugs, tricyclic antidepressants, the selective serotonin/norepinephrine reuptake inhibitors (SNRIs) duloxetine and venlafaxine, and opioids are used in the treatment of neuropathic pain.[11] Another effective drug for neuropathic pain is gabapentin. Gabapentin-mediated reduction of the symptoms of neuropathic pain is mediated by the inhibition of glutamate release in the spinal cord dorsal horn.[12]

Peripheral neurostimulation is also a treatment option for the treatment of these types of neuralgias. Rauchwerger et al[10] reported that they applied peripheral nerve stimulation to the patients that are refractory to pharmacologic intervention and had good results in controlling the pain. Rozen et al[13] reported the results of pulsed radio frequency in five cases who suffered from chronic inguinal

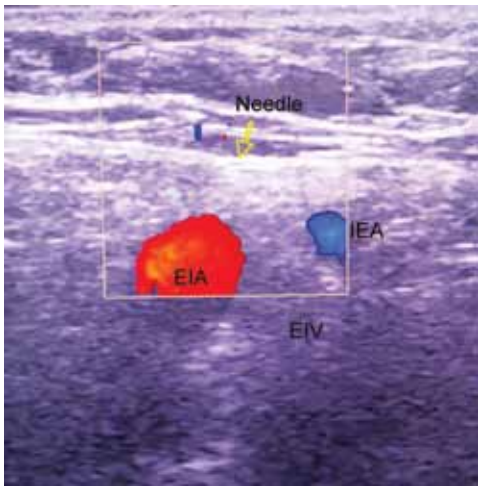

Fig. 2: Before local anesthetic injection

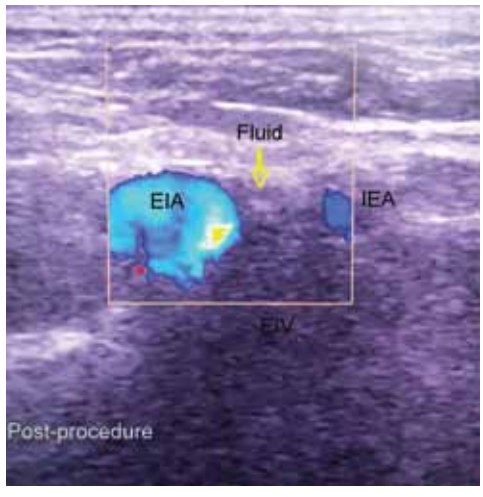

Fig. 3: After local anesthetic injection

pain. They applied pulsed radiofrequency after initial positive response to T12, L1 and L2 nerve blocks. All patients reported 75 to 100% pain relief lasting from 6 to 9 months. Fanelli et al[14] reported the results of cryoanalgesic ablation treatment in 10 patients with ilioinguinal (4 patients), genitofemoral (1 patient) or combined (5 patients) neuralgia. They applied 12 ablation procedures. Their mean follow-up period was 8.2 months.

Neurectomy and neural block are interventional treatment modalities in genitofemoral and ilioinguinal neuralgias. Feridun Acar et al[15] reported the highest documented number of patients with intractable ilioinguinal and genitofemoral neuralgia treated with neural block in the literature. Among their 20 patients with medi-cally intractable genitofemoral or ilioinguinal neuralgia, all patients received either nerve block or surgery and the mean follow-up was 11 months. In eight patients with one nerve block application, VAS score were 0 after 12 months. In four patients (two and in two patients), three nerve block sessions were applied at 1 week time intervals and their VAS score were 2 after 12 months. Six of the patients whose pain had not improved with nerve block application underwent neurectomy surgery which resulted in pain relief.

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