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## Scientific letter

# Laparoscopic neurolisys for the pudendal neuralgia treatment<sup>☆</sup>



## Neurólisis laparoscópica para el tratamiento de la neuralgia del pudendo

Pudendal neuralgia (PN) is an underdiagnosed chronic, disabling condition.

The diagnosis is not always straightforward, as the pelvic region is an area of confluence of several nerve trunks. Its difficult diagnosis and the fact that it is a relatively rare pathology often lead to a late diagnosis.

This condition affects women more frequently, and the average time for diagnosis is about 4 years, after a succession of consultations with different specialists.<sup>1</sup> The International Pudendal Neuropathy Association estimates an incidence of 1/100 000, although this number could be significantly higher.

The pudendal nerve originates from the S2, S3, and S4 roots. It is a motor, autonomic and sensitive nerve responsible for the sensitivity of the perianal and genital region as well as the motor innervation of the anal and bladder sphincters. It has 3 branches: perineal, rectal and distal, which is the dorsal of the penis in men and the dorsal of the clitoris in women.

PN is also known as pudendal nerve entrapment syndrome. It is produced by compression of the pudendal nerve along its pathway and at the exit or entrance of the pelvis by any of the surrounding osteo-musculotendinous or vascular structures.<sup>2</sup>

PN can be due to prolonged compression (cyclists or professional drivers), secondary to previous surgeries, vascular compression, or endometriosis. In 70% of patients, the nerve is trapped in the 'clamp' formed by the sacrospinous and sacrotuberous ligaments, in 18% it is entrapped in Alcock's canal, and in 10% the nerve rides on the falciform process of the sacrotuberous ligament.

Symptoms are defined by the Nantes criteria (Table 1), described by Labat in 2008<sup>3</sup>: pain in the perineal or perianal region that worsens upon sitting, is relieved when standing or lying down, and does not interrupt sleep.<sup>1,2</sup> CT-guided pudendal nerve block confirms diagnosis.<sup>3</sup>

We present our experience in laparoscopic pudendal neurolisis, with a description of the technique and its results.

Once the diagnosis is confirmed in our Pudendal Nerve Unit, the patients are treated with neuromodulators and epidural intracanal pulsed radiofrequency.

When the response to radiofrequency is poor, pudendal nerve decompression surgery is indicated.

We use the laparoscopic approach (4 trocars, patient in Trendelenburg) and access the obturator foramen through an incision to the peritoneum between the common iliac vein and the umbilical artery. Progress is made caudally by identifying the obturator nerve and obturator vein, and medially by rejecting the surrounding fatty tissue. The tendinous arch and the ischial spine are viewed and, after dividing fibers of the coccygeal muscle, the sacrospinous ligament is identified, which inserts into the ischial spine and extends medially. Below the ligament and close to the spine, we then identify the pudendal neurovascular bundle.

The entire ligament is divided (Fig. 1) until fatty tissue is reached, which indicates the entry into the Alcock canal, and the mobilization of the pudendal nerve is verified.

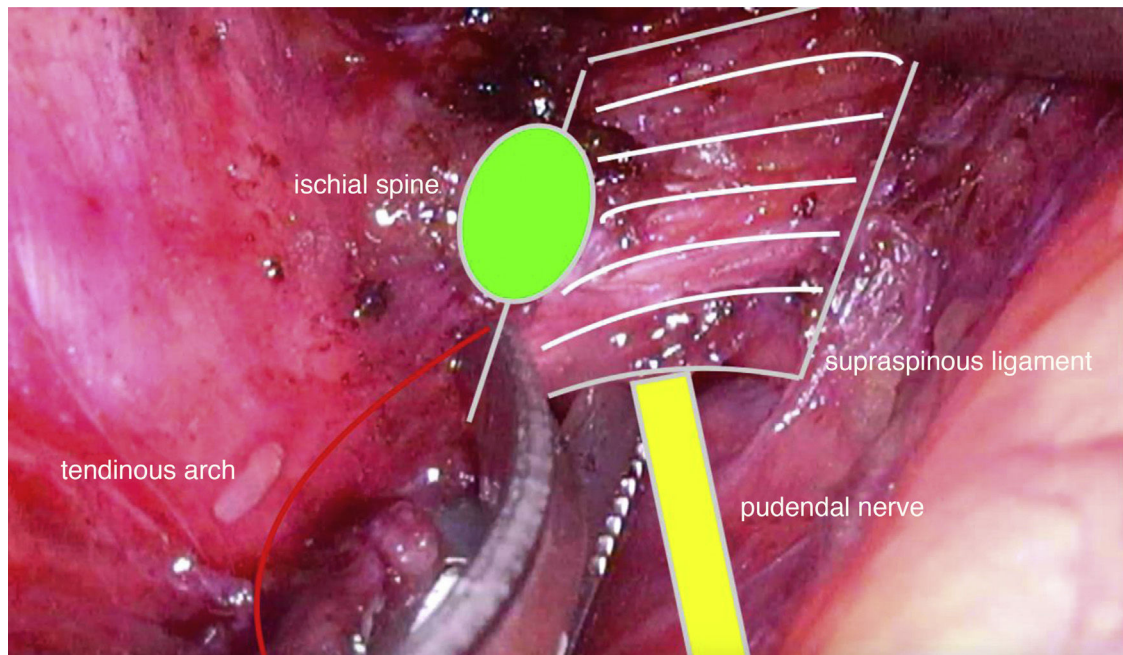
Surgical treatment has been shown to be the most effective and definitive in the management of PN with intractable pain.

At least 5 approaches have been published in the literature.

**Table 1 – Nantes major criteria.**

|   |
|---|
| Anal, perianal or vulvar pain                                     |
| Pain that becomes worse when sitting                              |
| Pain that is relieved in decubitus, allowing the patient to sleep |
| Pain without sensory impairment                                   |
| Pain relieved by diagnostic pudendal nerve block                  |

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**Figure 1 – Division of the sacrospinous ligament.**

The transperineal approach was described by Shafik in 1991<sup>4</sup> and the transvaginal approach by Baurant in 2003<sup>5</sup>; both techniques are practically blind and very personal. These authors report success rates between 62% and 70%.

The transgluteal approach was described by Robert, and it is the only approach supported to date by randomized studies<sup>6</sup> with a success rate of 71%. Technically, it is very demanding, limiting its action to ligamentous impingement, and potential sequelae include fibrosis and persistent pain.

The robotic approach shares the same access as the laparoscopic technique and improves it in terms of image and dexterity.

The laparoscopic approach, described by Possover in 2004,<sup>7</sup> provides an excellent view of the nerve plexuses with great definition. It therefore provides the ability to simultaneously diagnose and resolve other causes of neurological compression, such as endometriosis and postoperative fibrosis. In a series of 134 patients, Possover achieved improvement in 83% of patients.

Between March 2018 and December 2019, we performed 6 laparoscopic pudendal neurolysis procedures in 5 patients. Four patients underwent unilateral neurolysis and one patient a bilateral procedure in 2 independent interventions. All patients were female, and the mean duration of their symptoms was 4 years (2–8).

The mean operative time was 65 min (40–80). The postoperative stay was between 24 and 72 h. We had no relevant complications.

Pain intensity was measured by the visual-analog scale before surgery, in the postoperative period, and after 6 months.

In 5 out of the 6 procedures, the patients presented a reduction in pain intensity of at least 50%, which was maintained 6 months later.

In our experience, laparoscopic neurolysis of the pudendal nerve has been shown to be a minimally invasive technique that is technically feasible and safe. It provides for excellent exploration of the neurological structures of the presacral and obturator regions, as well as precise dissection of the pudendal nerve.

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## Calcifying nested stromal-epithelial tumor: A rare hepatic neoplasm<sup>☆</sup>



### Tumor calcificante en nidos epitelial-estromal: neoplasia hepática excepcional

Calcifying nested stromal-epithelial tumors (CNSET) are rare primary liver tumors<sup>1-6</sup> that were first described by Ishak in 2001<sup>1,3,4,6</sup>. Only 43 cases have been published in the literature<sup>2-9</sup> (Table 1). These neoplasms present an apparently benign and indolent clinical course, attributable to their low malignant potential<sup>10</sup>. Surgical treatment with free margins usually provides high long-term survival rates.

We present a 21-year-old patient with no relevant history. Follow-up lab work showed an altered liver profile (gamma glutamyl transpeptidase [GGT] 122 IU/L). Physical examination revealed hepatomegaly. Abdominal ultrasound identified a hyper-isoechoic focal lesion, apparently calcified, with a lobulated shape and limited vascularization. Magnetic resonance imaging (MRI) and computed tomography (CT) scan (Fig. 1A-B) showed a large, irregular, heterogeneous mass with multiple calcifications, with irregular enhancement in the arterial phase and washout in the portal phase. The initial diagnosis was fibrolamellar hepatocellular carcinoma (FHCC) in segments IV, V, VI, VII and VIII. Tumor marker levels (Alpha-fetoprotein, carcinoembryonic antigen and CA 19.9) were normal. Total liver volume was 1172.83 cc. The calculated residual volume percentages were: 42.2% for segments II-III (495.59 cc) and 48% for segments I-II-III (562 cc). Due to the suspicion of FHCC, surgery was indicated, and we performed right trisectionectomy by laparotomy as well as lymphadenectomy of the hepatic hilum and celiac trunk. The postoperative period transpired without incident, and the patient was discharged on the 7th postoperative day.

The pathological study reported a whitish nodular tumor measuring 21 × 13 × 8 cm with multiple calcifications, areas of

ossification and lymphadenopathies without neoplasm. Immunohistochemistry showed the cells were positive for Vimentin, Actin, WT-1, CD56, CD99, CD117 with Ki67:1% (Fig. 1C-D). These results were compatible with CNSET with free surgical margin, no perineural invasion or lymphovascular permeation.

The case was presented to the Oncology Committee, who decided to monitor the patient in an outpatient clinic without adjuvant chemotherapy because there is no clear benefit to justify adjuvant therapy. Six months after surgery, the patient remains asymptomatic and disease free. Follow-up studies included laboratory testing with liver panel and CT scan.

CNSET have been described in the literature with various terminologies: ossifying malignant mixed epithelial and stromal tumor, ossifying stromal epithelial tumor, and desmoplastic nested spindle-cell tumor of the liver<sup>1</sup>. It was Markhoulf who proposed the term CNSET because it incorporates all the characteristics of the tumor.

Despite their exceptional nature, we know that they present more frequently in young (<20 years) or pediatric patients (77%) and females (70%) (Table 1). They are usually located in the right hepatic lobe (64%), and in 40% of cases their diagnosis is incidental (Table 1). The association of CNSET with hormonal alterations is notable (36%): Cushing syndrome or cushingoid symptoms (25%)<sup>5,7</sup>, Klinefelter syndrome<sup>4</sup> and oral contraceptives. Some authors have tried to link the development of these tumors with hormonal alterations, but this has not been proven. In four patients, it was associated with Beckwith-Wiedemann syndrome (overgrowth syndrome with increased risk of developing cancer)<sup>2,8</sup>. Our case did not present an association with these pathologies.

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