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Case Report

Osteopathic manipulative treatment in pudendal neuralgia: A case report

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ABSTRACT

Pudendal neuralgia is characterised by pain in the pudendal dermatome. It could be due to a stenosis of the pudendal canal, a compression along its pathway, or a pelvic trauma.

Pudendal nerve entrapment (PNE) syndrome is frequently involved in pudendal neuralgia onset. This case report describes the osteopathic manipulative treatment (OMT) of a patient with functional PNE. A 40-year-old female presented with a 12-month history of intense pelvic pain resistant to 3 months of pharmacologic treatment that arose after three proctological surgeries. A perineal retracted painful scar was visible upon examination. PNE syndrome diagnosis was based on Nantes criteria. The electromyogram of the nerve showed an increased motor response latency of the left pudendal nerve. Visual analogue scale (VAS), female National Institutes of Health Chronic Prostatitis Symptom Index (NIH-CPSI), Oswestry Disability Index (ODI) and Tampa scale of kinesiophobia (TSK) were used to assess patient's symptoms at baseline (T0), after pharmacologic treatment (T1), after OMT (T2), and at 6-month follow-up. Five treatments, including direct and indirect techniques, were performed over 1 month. OMT reduced pelvic neuralgia and disability indexes without any complications, maintaining a positive outcome at 6-month follow-up (VAS: T0 = 10, T1 = 10, T2 = 1.8, T3 = 1.5), (NIH-CPSI: T0 = 34, T1 = 30, T2 = 7, T3 = 6), (ODI: T0 = 48, T1 = 29, T2 = 9, T3 = 5) and (TSK: T0 = 51, T1 = 41, T2 = 20, T3 = 17). This is the first report of a patient diagnosed with functional PNE managed with OMT. A link between PNE, scar and pelvic somatic dysfunctions could suggest double crush syndrome.

1. Introduction

Pudendal neuralgia is a debilitating condition, predominant in females; it consists of perineal pain along the pudendal nerve territory that is caused by inflammation, compression or entrapment in the Alcock's canal, which are recognised as its common aetiology (Labat et al., 2008). Pudendal neuralgia prevalence is estimated to be 1% in the general population, reaching a substantially higher percentage in post-abdomino-pelvic surgery patients (Pailhé et al., 2013; Possover and Forman, 2015; Ploteau et al., 2016). Other associated exposing factors are childbirth, intense cycling and age-related changes. The diagnosis of PNE is clinical, based on the Nantes criteria, edited in 2008, as imaging and laboratory diagnostics are not reliable enough in this condition. Notwithstanding this limitation, warning signs that should suggest specific investigations include excessive neuropathic pain, pain waking the patient at night, pinpointed pain, and severe pelvic motor deficits (Labat et al., 2008; Ploteau et al., 2016). Pudendal nerve entrapment (PNE) syndrome aetiology is the entrapment of the pudendal nerve along its course, most often in three main areas: between the sacrotuberal and sacrospinous ligaments, within the pudendal canal, and over the sacrotuberal ligament. Neurophysiologic investigations such as electromyography should be performed to better study the peripheral nervous system functionality (Lefaucheur et al., 2007). Additional causes of pudendal neuralgia, which should be considered in the differential diagnosis, include pudendal neuroma, sacroiliac skeletal abnormalities, pelvic trauma, ganglion cyst, and rectal cancer (Capek et al., 2014; Pérez-López and Hita-Contreras, 2014).

In this case report, we documented pudendal neuralgia in a proctological postoperative patient, which was treated with osteopathic manipulative treatment (OMT).

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2. Case report

A 40-year-old female, nullipara, hospital employee, with normal body mass index, was referred to the osteopathic outpatient clinic after she reported intense constant perineal burning pain, spreading from the left side of the perineum to the genital area, associated with vulvar pain, dyspareunia, and perineal numbness. Defecation and seated position exacerbated the pain, but the latter did not wake the patient at night. No pelvic organ dysfunctions such as urinary incontinence were revealed. The patient also mentioned pericoccygeal pain, which worsened with the sitting position but was not always concomitant with the perineal pain. The patient's pathologic history was negative for gastrointestinal or gynaecologic inflammatory diseases, and a surgical treatment for a pyloric stenosis was reported in her clinical past. In March 2012, the patient underwent haemorrhoidectomy for a symptomatic grade III setting. Six months later, a relapse occurred, which required a second haemorrhoidectomy. Five months after the second surgical treatment, an anal fissure degenerated into an ano-rectal fistula, which exposed the patient to frequent infections; in March 2013, a lateral internal sphincterotomy was performed. Related to this, a painful hypertrophic retracted scar was detected on the left posterior quadrant upon examination. Two weeks after surgery, pudendal neuralgia started, which intensified in the subsequent months. The symptoms were not present before the last ano-rectal surgery. Hence, the patient visited a neurologist, who diagnosed PNE syndrome, after excluding other possible pathologies with the contribution of previous colonoscopy and proctoscopy, which were both negative. Other possible pathologic implications were ruled out through routine proctological evaluation. A motor electroneurography of the pudendal nerve was performed using a St. Mark pudendal electrode, which showed a diminished amplitude in the response of the left nerve, probably due to its entrapment inside Alcock's canal. Specifically, the pain timing and symptoms fulfilling Nantes criteria suggested nerve sprain. A 3-month pharmacologic treatment with anticonvulsants and an analgesic was proposed (Pregabalin, Duloxetine, and transdermal Fentanyl). The physician suspected the relationship between pain and pelvic girdle dysfunction relying on the pudendal sprain objectivity and the patient's multi-traumatic history, also in the light of a retracted scar discovered during the clinical assessment; the physician then opted for a functional therapy such as OMT in the case of persistent symptoms. During the first osteopathic assessment, pain and disability were evaluated, defining the (T1) along the timeline (see Table 1).

Physical examination was performed to qualify the clinical picture and somatic dysfunction relevance. Subsequently, a specific osteopathic assessment was performed (pelvic girdle joints motion tests, sacral bases rocking test, fascial assessment of thoracic diaphragm and pelvic floor, and palpation of epi-cranic fascia). The palpation of left sacrospinous, sacrotuberous, sacroiliac, and lumbo-sacral ligaments elicited pain, while tenderness was revealed with the palpation of left gluteal muscles and pelvic floor muscles. Prone lumbar instability and active straight-leg rise were negative. Gillet and the posterior pelvic pain provocation tests were positive on the left side. Tenderness and elicited pain manifested on the symptom provocation test of the sacro-coccygeal joint, together with the absence of motion during the respiratory motion test (Origo et al., 2017). OMT was performed due to the evidence of dysfunctional pathways in the clinical picture.

The patient's outcome evaluation was performed using outcome scales and pain index. Visual analogue scale (VAS), female National Institutes of Health Chronic Prostatitis Symptom Index (NIH-CPSI), Oswestry Disability Index (ODI) and Tampa scale of kinesiophobia (TSK) were used to assess pelvic pain, disability and movement fear. The assessment was carried out four times at the following time durations:

- T0: The baseline evaluation made by the physician during the first visit
- T1: Evaluation of symptoms after 3 months of pharmacologic treatment
- T2: Early follow-up after 1 month of OMT
- T3: 6-month follow-up, starting from the last treatment.

In detail, NIH-CPSI and VAS were used to be more specific for pudendal neuralgia, while ODI and TSK were chosen to estimate disability and kinesiophobia related to pelvic girdle pain, assuming a functional relationship between pudendal pain and pelvic girdle disability (Clemens et al., 2009; Vleeming et al., 2008).

The OMT lasted 1 month with the administration of five treatments (one per week), combining the use of direct and indirect fascial techniques; these were selected after physical examination and osteopathic differential analysis, specifically focusing the attention on the double crush syndrome (DCS) mechanisms by presuming an irritant role of the pudendal canal and the dura mater in the region of emergence of the sacral roots (S2-S3-S4). The osteopathic techniques used in the first two treatments were pelvic floor release, balanced ligamentous tension technique of the sacroiliac joints, fascial unwinding technique of the hypogastric region, lumbar-sacral decompression, and internal direct sacro-coccygeal mobilisation (Maigne et al., 2006). The last three treatments included fascial unwinding of the thoracic diaphragm, the pelvic floor, and the crural fascia. Furthermore, the balanced ligamentous tension technique of the sacrotuberous ligaments was carried out in combination with the tension balancing of the extracranial fascia and the dura mater for both the skull base and the sacrum (Tozzi, 2012). The number of treatments performed was based on the clinical experience and patient's pain course, presuming a maximum medical improvement after five treatments.

The baseline evaluation of the patient manifested severe disability and high pain levels, as attested with outcome scales. After 3 months of pharmacologic treatment, the second assessment showed a mild improvement in disability, from severe to moderate in (T0) (VAS = 10, NIH-CPSI = 34, ODI = 48, TSK = 51), and a substantially unchanged pain perception in (T1) (VAS = 10, NIH-CPSI = 30, ODI = 29, TSK = 41). Early follow-up was performed after the five OMT sessions (T2) (VAS = 1.8, NIH-CPSI = 7, ODI = 9, TSK = 20). Improvement of the pudendal neuralgia and associated disability was achieved, with a substantial improvement of the patient's activities of daily living, as a minimal disability score level was reached. Functional and symptom provocation tests were negative at the end of the treatments and at follow-up. At 6-month follow-up (T3), the outcomes confirmed a stable progressive reduction of disability and fear of movement, conducive to a full recovery of the patient's functionality (VAS = 1.5, NIH-CPSI = 6, ODI = 5, TSK = 17) (see Table 2).

Table 1
Timeline tracking the patient's history.

Date	Assessments	Procedure
March 2012		1st Haemorrhoidectomy
September 2012		2nd Haemorrhoidectomy
March 2013		Lateral internal sphincterotomy
May 2013	T0	Baseline assessment
May 2013		Start of pharmacologic therapy
August 2013	T1	End of pharmacologic therapy
September 2013		Start of OMT
October 2013	T2	End of OMT
April 2014	T3	6-month follow-up

Table 2
Outcome scale values according to assessment time points. A significant decrease in disability can be observed at (T2) evaluation after five OMTs.

	T0	T1	T2	T3
VAS	10	10	1.8	1.5
ODI	48	29	9	5
NHI-CPSI	34	30	7	6
TSK	51	41	20	17

3. Discussion

Pudendal neuralgia is a multifactor process with physiologic and psychological implications. Anxiety is considered a sizable condition for interpretation of the case and its clinical course (Renzi and Pescatori, 2000). The Nantes criteria can be used to diagnose PNE by using five main criteria and eight secondary criteria, while imaging and other laboratory tests cannot reach sufficient reliability and specificity. In this case, the patient presented all Nantes criteria, except for the last one, as a pudendal nerve block was not performed. Coccydynia and PNE association is not common, but in this case, through physical and osteopathic examination, a link between PNE, coccydynia and somatic dysfunctions emerged, which suggested DCS. The patient followed medical therapy with slight but not sufficiently effective improvement of disability; even if the pharmacologic treatment was required, it was not resolutive for both pain and disability. On the contrary, after 1 month from OMT start, symptoms and disability were relieved, and the patient recovered full activities of daily living. What emerged from the 6-month follow-up was the maintenance of the results, reaching a stable clinical condition (see Fig. 1).

Pelvic, uro-genital and colorectal surgeries are exposing factors for the onset of pudendal neuralgia, probably due to abdominopelvic peritoneal adherences and scar formation, which compromise the fascial layer mobility, hence triggering pro-inflammatory activity (Venturi et al., 2015). The patient's response to the two distinct treatments (pharmacologic and OMT) could be determined by the functional involvement of the elasticity of the perineal structures, not only concerning the regional fascia, but also concerning a fascial network based on anatomic connections (pelvic girdle fascia, dura mater and crural fascia) (Willard et al., 2012). Kajita et al. (2013) observed that the presence of a painful scar in somatic nerve territory can make it more sensitive and predisposed to peri-neural tissue inflammation and to a lower pain threshold of the nerve involved. Furthermore, a distinct compression at two or more locations along the course of a peripheral nerve can coexist and synergistically increase symptom intensity (Kane et al., 2015). We hypothesised an overlapping of DCS and pudendal nerve entrapment, which worsened the patient's condition. In this specific case, during osteopathic assessment, pelvic girdle dysfunctional pathways and abnormal fascial tension were identified between two anatomical landmarks: the first one in the sacro-coccygeal region (near the insertion of the dura mater, the filum terminale) and the second one at the level of the pelvic scar (Lo et al., 2012; Cohen et al., 2016). In a previous study performed on patients with coccydynia, the administration of OMT in patients complaining of radicular pain associated with coccydynia showed effects not only on coccygeal pain but also on the coexisting radicular pain (Origo et al., 2017). However, no specific evidence of DCS on PNE syndrome exists, because many of the studies investigated the DCS on the upper extremities. The presence of pudendal nerve entrapment, together with the multiple pelvic trauma history, is suggestive for the concomitant existence of functional DCS in this patient. Further research on load transfer and pelvic nerve fibrous connective tissue changes is needed to understand the role of pelvic dysfunctions, including the possible effects of OMT.

With regard to DCS, there is still lack of well-established diagnostic criteria; the diagnosis remains clinical based, which poses a limit for clinical and anatomopathological estimation of the case progression.

In summary, painful scar, DCS and somatic dysfunctions hypothetically led to the lessening of connective tissue elasticity and to the upholding of the pudendal neuralgia, resistant to pharmacologic treatment (see Fig. 1).

4. Conclusion

This paper reports the first case in literature of OMT management of functional PNE syndrome. OMT was demonstrated to be effective in this patient's symptoms remission and disability reduction, as evaluated using multiple outcome scales. Nevertheless, this clinical hypothesis should be tested with appropriate studies to determine long-term and short-term effects of OMT on

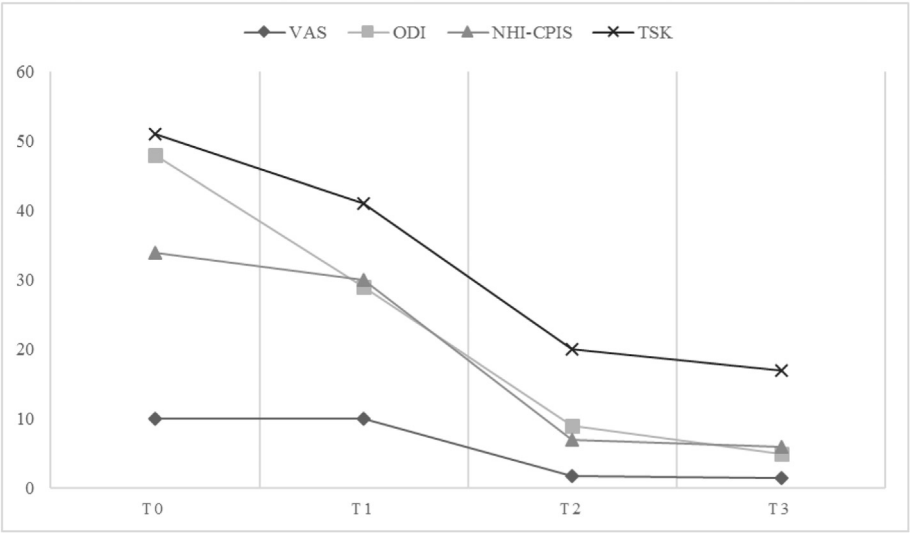

Fig. 1. The Y axis shows scale values, while the X axis indicates assessment time points. The line chart highlights the clinical condition improvement, especially among (T1) and (T2).

chronic pelvic pain and pudendal neuralgia, led by dysfunctional pathway.

Consent

Written informed consent was obtained from the patient for publication of this case report.

Disclosure and conflicts of interest

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors. The authors declare they have no conflicts of interest.

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