Original article

Differential staged sacral reflexes allow a localization of pudendal neuralgia

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Abstract: The objective of this work is to localize pudendal nerve compression by measuring sacral reflexes and to explain differences in latencies by an anatomical investigation. Electrophysiological data was obtained from 59 patients by measuring the healthy side and the painfull side of 79 patients with uni- or bilateral pain. In addition, 7 formalin-fixed female cadavers were dissected to identify nerve trajectories. Dissections demonstrated that the levator ani nerve runs directly from the sacral plexus, to innervate the levator ani, while the pudendal nerve traverses the sacrospinal and sacrotuberal ligaments, entering the perineum with an inferior rectal nerve, already separated runs to the posterior or caudal part of the external anal sphincter muscle, while the anterior parts of the external anal sphincter muscle are innervated by perineal nerve branches coming from the pudendal canal. Significant differences in latencies suggest that main nerve compression occurs in the pudendal canal. In conclusion, it is possible to localize differences in pudendal nerve compression by separate electrophysiological examination of staged sacral reflexes and allows to narrow down the location of nerve compression.

Key words: Anatomy; Compressive neuropathy; Electrophysiology; External anal sphincter innervation; Pudendal nerve.

INTRODUCTION

Compressive pudendal neuropathy is a frequent condition that is often ignored.[1-3] Its incidence is approximately 1% in the general population and the condition probably affects women more often than men. Because healthcare professionals lack an adequate method to diagnose accurately pudendal neuropathy, the affected individuals often embark on an endless quest for effective relief with serious physical and psychological consequences.

There are several possibilities of a treatment to diminish pain. The first line is conservatory followed by infiltrations [4, 5] and, ultimately by surgical procedures.[6-9] It is therefore important to identify precisely the site where the nerve is compressed. The classical approach consists of measuring pudendal nerve terminal motor latencies after electrophysiological stimulation. However, the results obtained with this technique lack of reproducibility and sensitivity.[10-14] The endo-vaginal or endo-rectal stimulation at the level of the ischial spine have been criticized because of poor precision in the localization of the stimulation point, the distortion of the stimulation potential by the different intervening tissues and due to the fact, that albeit an increased latency did demonstrate myelinopathy, it could not indicate its location. Several conditions may interfere with conduction times, including vascular factors, presence of distal synaptic ends, vegetative reactivity and time-dependent variability.[15-17] Moreover, a compression at the level of the ischial spine or just beside it cannot be detected by pudendal nerve terminal motor latencies. Thus, the sacral reflex measurements were thought to be a good indicator for the extent of the affected terminal nerve area. However, it is usually recorded via the bulbocavernosus muscle; yet it reveals to be unsatisfactory.[18] Therefore, the aim of our study was the development of a more precise diagnostic test to localize pudendal neuropathies. Preliminary electrophysiological tests of the anterior and posterior innervations in the anal sphincter area (Eric de Bisschop, unpublished results), revealed marked latency differences. Therefore, we decided to compare sacral reflex transmission latencies through the pubococcygeus loop and the levator ani and the anterior and posterior anal sphincter parts, calling them *"staged sacral" reflexes*. Differences in latencies due to electrophysiological measurements were validated and so it can be explained by differential innervation of the anal sphincters by rectal inferior and perineal nerve branches.

SACRAL REFLEXES

When a nerve is compressed, its vascularization is compromised and consequently the nerve suffers. Nerve suffering has several consequences, including a decrease in nerve conduction velocity that affects both the motor fraction of the sacral reflex and the sensory transmission owing to the evoked somesthesic potentials. This decrease can be recorded as a prolonged latency period between the stimulus and the record points. In a pudendal nerve terminal motor latency measurement, the stimulation electrode is placed over the ischial spine usually by endo-rectal insertion. An impulse is delivered and a recording is made using a surface electrode placed on the perineal muscle, usually at the level of the anal sphincter (Fig. 1A). The stimulus may also be delivered through an endo-vaginal insertion. In both cases, the examination consists of measuring the latency, i.e. the time elapsed between the moment of impulse delivery and the record of the electric potential in the target region. By comparing the recorded latencies to normal values, it is possible to estimate the influx transmission capacity of the nerve. Normal latencies, reported in the literature, vary from 2 to 5.35 msec.[14, 19] The measurement of the sacral reflex is carried out in an analogous manner: a stimulus is delivered at one point and the potential is recorded at a second point. The stimulus is delivered in this case at the level of the clitoris. The impulse travels through the sensory pudendal nerves towards the sacral spine (Onuf's nucleus) and returns back through motor efferent nerves. The potential is recorded in one of the zones of the pudendal nerve that is innervated by motor fibers. In case of nerve compression, latency is increased due to the impairment of nerve conduction (Fig. 1B).

CONVENTIONAL ANATOMY

In a conventional anatomical description, the pudendal nerve originates as a fusion of the S2, S3 and S4 roots, with an occasional involvement of the S1 and L5 roots, while the levator ani muscle is innervated by a nerve that originates

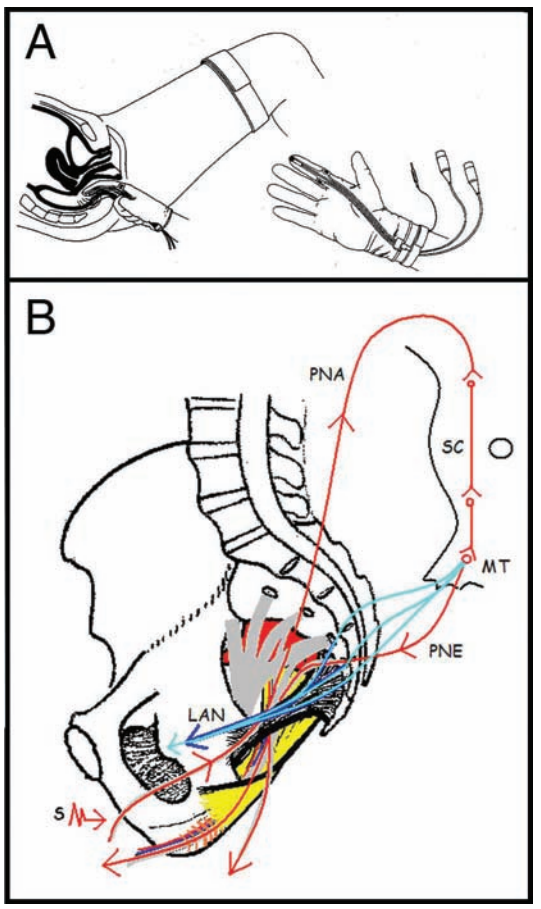

Fig. 1. – (A) The technique of pudendal nerve terminal motor latency measurements. (B) Electrophysiological Anatomy. S: indicates the stimulus at the level of the clitoris. Records are made at the level of the anterior and posterior anal sphincter quadrants. LAN: levator ani nerve. PNA: pudendal nerve afferents. PNE: pudendal nerve efferents. MT: medullar transfer. SC: spinal cord.

in the sacral plexus and runs above the levator ani muscle (on top of the pelvic floor) on the pelvic side and then innervates the iliococcygeus and pubococcygeus muscles as well as the upper part of the puborectalis.[18-20] The pudendal nerve circumvents or perforates the sacrospinous ligament (formerly called small sacro-sciatic ligament) and enters between sacrospinous and sacrotuberous ligaments (formerly also called great sacro-sciatic ligament) into the perineum, also called "ligament clamp" as it constitutes a site for potential pudendal nerve compression. Nerve and blood vessels run within the ischiorectal fossa towards the anterior perineum and vasculo-nervous elements enter the pudendal canal (also termed Alcock's canal), that is constituted by duplication of the fascia of the internal obturator muscle, thus may also cause a nerve compression. The inferior rectal nerve classically innervates all parts of the anal sphincter, and separates from the pudendal nerve at the beginning of the pudendal canal and runs across the ischiorectal fossa towards the anal sphincter. In the middle of the pudendal canal, the pudendal nerve divides into two branches: the dorsal nerve of the clitoris and the perineal nerve, with both nerves traversing the pudendal canal in its entirety. The perineal nerves give off the sensory branches to the perineum as well as the motor branches for the perineal muscles and for the external anal sphincter. The dorsal nerve of the clitoris is a terminal sensory branch of the pudendal nerve.

Here, we confirm that the anterior and posterior quadrants of the anal sphincter are innervated by different branches and by different trajectories and provide several possible entrapments of parts of the nerves and consequently pudendal neuropathy. Because the afferent path of the dorsal nerve of the clitoris is always the same, it is theoretically possible to locate the site of compression with some precision by examining the different efferent paths. To confirm the validity of this assertion, we carried out an anatomical study aimed at determining:

i) the systematic presence of a nerve in the levator ani (pubococcygeus) muscles;

ii) the location of the dorsal nerve of the clitoris starting point relative to the main trunk;

iii) the presence of a starting point of the inferior rectal nerve that would be more proximal than what is described in the literature;

iv) to identify differences in the innervation of the anterior and posterior anal sphincter quadrants.

MATERIALS AND METHODS

We relied on 27 consecutive male and 49 female patients suffering from unilateral or bi-lateral pudendal neuropathy. The diagnosis was ascertained because symptoms disappeared after conservative treatment or surgery. We hypothesize that the absence of symptoms on the controlateral part was the guaranty of an intact pudendal anatomy. Also for ethical reasons we decided to use each patient as his own comparative and the healthy side as control value. Each patient had 3 measurements on each side meaning 6 values at all. The first record was the reference for the pubo-rectal muscle, the second was taken at the superior quadrant of the external anal sphincter and the third at the inferior quadrant of the external anal sphincter. The obtained values show that nerves may be affected differentially and exhibiting significantly higher electrophysiological values on the affected side than on the normal side. To ascertain that our hypothesis was correct, we decided to confirm the anatomical situation on cadavers. The seven corpes used in this study were obtained by the donation program to the Department of Cellular Biology and Morphology. All donors gave previously a written consent. The cadavers were perfused through the femoral artery with a mixture of 0.9 L of formaldehyde (38%), 0.5 L of phenol (85%), 1.0 L of glycerol (85%), 4.0 L of ethanol (94%) and 10.6 L of water. The cadavers, were stored at 8 °C until dissected by second year medical students as part of their training. Half pelvises that had not been dissected in the course were used to study the trajectories of the anal, pudendal and perineal nerves described in this work.

RESULTS

The nerve fibers for levator ani (pubococcygeus) muscle run directly from the sacral plexus, above the pelvic floor on the side of the levator ani and follow a different direction from the pudendal nerve (Figs. 2A and 2B). The pudendal nerve traverses the ligament clamp located between the sacrospinal and the sacrotuberal ligament in the lateral space of the ischiorectal fossa where nerves and vessels are surrounded by the fascia of the internal obturator muscle and form the pudendal canal. In one of the seven dissections performed, the pudendal nerve actually perforates the sacrospinal ligament. In five cases, the pudendal nerve divides into the dorsal nerve of the clitoris and perineal nerves after passing underneath the sacrospinal ligament but before entering the pudendal canal (Fig. 2C). The same anatomical section also shows that the dorsal nerve of the clitoris runs parallel to the pudendal canal. The neurovascular bundle within the pudendal canal gives rise to multiple perineal branches that branch off either at the point of entry into the ischiorectal

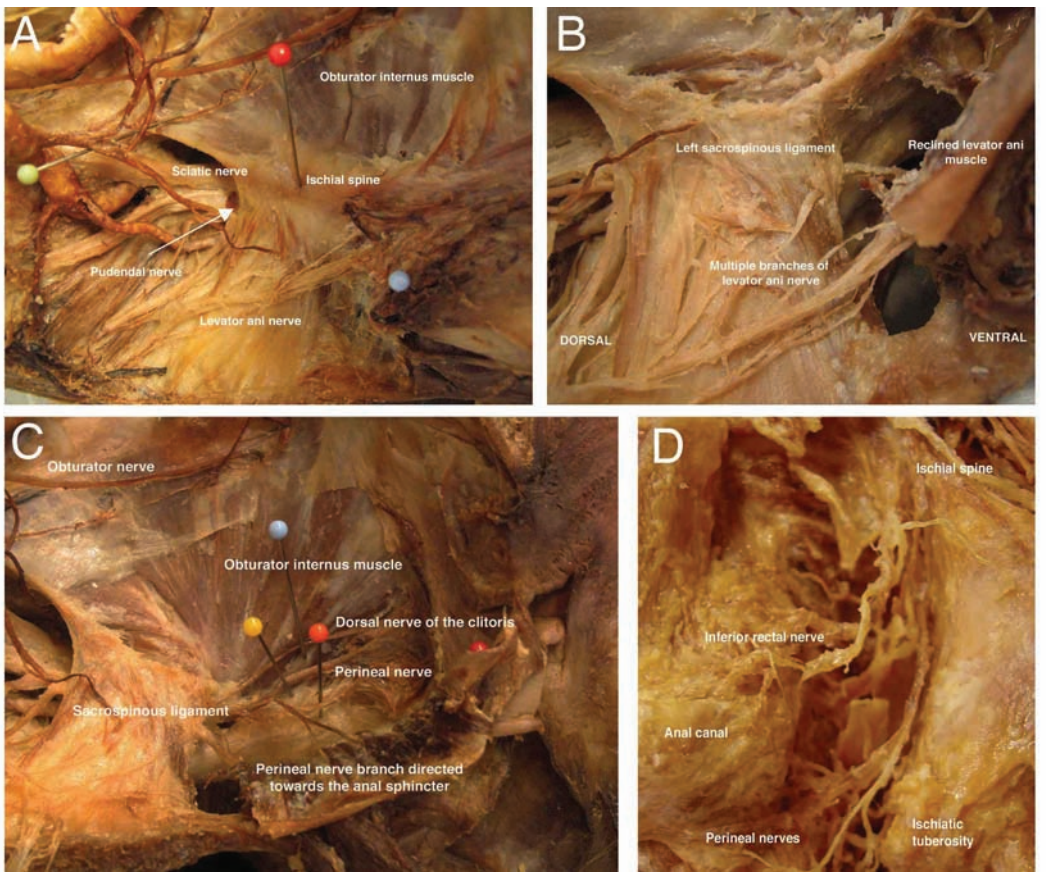

Fig. 2. – (A) The internal face of the left half pelvis. The red needle indicates the position of the ischial spine: the levator ani nerve passage through the sacrospinal ligament is clearly visible. (B) The passage of the levator ani nerve above the sacrospinal ligament is easily distinguishable. (C) The pudendal canal has been opened and the internal obturator fascia removed. The nerve that passes above the blue needle is the dorsal nerve of the clitoris, the one above the yellow needle is the main branch of the pudendal nerve, i.e. the perineal nerve. The ramifications of the inferior rectal nerve are seen underneath the red needle. (D) Ischiorectal fossa in the right half pelvis. The inferior rectal nerve that innervates the posterior anal hemisphere originates in a much higher zone and does not traverse the pudendal canal as classically described in the literature. In addition to other functions, perineal nerves act as motor nerves for the anterior anal hemisphere.

fossa, inside the pudendal canal in the direction of the anal canal (sphincter) or the anterior perineum. In all of the dissections, the inferior rectal nerve originates prior to the entry into the ischiorectal fossa and runs straight towards the back of the anal canal, through the ligament clamp but not through the pudendal canal (Figure 2D).

The electrophysiological measurements of the staged sacral reflexes show a significant higher latency time of the ventral quadrant of external anal sphincter innervation, while the posterior or caudal quadrant was just slightly prolonged and levator ani innervation was unaffected (Fig. 3A-C).

DISCUSSION

Historically, the pudendal nerve has been investigated using terminal motor latency,[14-21] a technique which is not very reliable.[22] Indeed, because of the distance between the stimulation point and the nerve (approximately 1 cm),[23] it is possible that the electrical impulse delivered at the point of contact with the ischial spine does not merely travel along the nerve but diffuses in the entire perineal region. Several investigators have made different contributions to the available tests.[24, 25] The levator ani nerve appears to be recognized as a distinct entity by all authors. Our work has shown that the levator ani is innervated by a nerve that originates in the sacral plexus. This nerve runs above the sacrospinal ligament and terminates in the pubococcygeus, the iliococcygeus, and the puborectalis muscles. This finding is supported by investigations carried out by Hallner[26] who failed to find any pudendal nerve innervation of these muscles in 200 dissections. At the same time, it should be noted that a few authors have nevertheless proposed possible pudendal nerve innervation of the levator ani muscle. In most cases, these opinions are based on experiments in which this nerve was anesthetized at the level of the ischial spine, resulting in the paralysis of the levator ani muscle.[27] The methodology of such experiments is however questionable: the anesthetic could have easily diffused towards the levator ani nerve, located less than 1 cm away from the anesthetic injection point. The dorsal nerve of the clitoris is classically described as a terminal branch of the pudendal nerve.[26] However, in our dissections it often appears as a branch that is parallel to the pudendal nerve and that does not run through the pudendal canal. This observation, which contradicts the classical description in literature, has also been confirmed by other authors.[7] If this observation is correct, then the dorsal nerve of the clitoris can hardly be compressed, which explains the interest in using this nerve as an afferent branch in sacral reflex studies. An even bigger controversy surrounds the inferior rectal nerve.[28-32] Certain authors believe that it originates at the beginning point of the pudendal canal,[33] with some variations.[32] Thus, its origin may be independent with or without an anastomotic branch from the perineal nerve. Moreover, the inferior rectal nerve may perforate the sacrospinal ligament in its middle portion at an approximate distance of 1 cm from the ischial spine, extending to a distance of up to 1.5 cm. Other investigators[33] estimate that 60% of

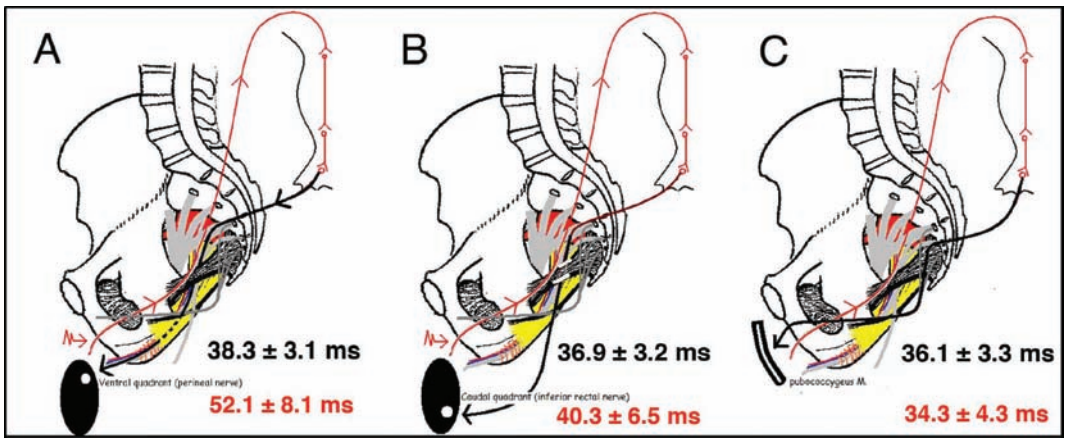

Fig. 3. – The concept of staged scared reflexes and its electrical impulse circuits. Image (A) represents the recording made from the anterior or ventral quadrant of the external anal sphincter. Image (B) represents the recording made from the posterior or caudal quadrant; and image (C) represents the recording from the levator ani (pubococcygeus) muscle. To establish normal latency of staged reflexes, the healthy side of patients with uni-lateral neuralgia were measured (n = 59), black numbers. Staged reflexes were measured in the painful side of 76 patients with uni- or bi-lateral neuralgia (numbers in red). Note the highly significant difference (t-test $p < 0.1$) of latencies of the anterior quadrant when compared with the healthy side, suggesting a compression of the pudendal nerve in the pudendal canal.

the lower rectal nerves originate from the pudendal canal. This view is supported Shafik and Doss that also describe an emergence from the pudendal canal. [31] In fact, it seems that any structure that terminates at the anal sphincter is referred to as an inferior rectal nerve. However, according to our findings, the anal sphincter shows at least two distinct innervations that may explain the differences with the classical descriptions in literature. The first innervation network originates from the inferior rectal nerve itself, while the second network is generated by the pudendal nerve via perineal branches. We believe that the multiple origins of innervation are important from both the clinical and electromyographical points of view (Fig. 3A-C). Indeed, one branch is the inferior rectal nerve that branches off very early (proximally) and runs parallel to the nerve, but is not located in the pudendal canal and no longer at risk for compression. This nerve terminates in the posterior hemisphincter. A second branch that innervates the anterior hemisphere appears to originate from a more distal region, i.e. the perineal branch of the pudendal nerve. It is thus subject to compression at the level of the ligament clamp and the pudendal canal or the falciform process. In other words, we believe that it is anatomically correct to postulate that the anterior and posterior anal sphincter quadrants as well as the pubococcygeus muscle all have separate and distinct innervations. As a consequence, a staged sacral reflex analysis of the reflex loops specific for these three muscles should make it possible to better locate the site of compression with greater precision compared to the current techniques. The anatomical analysis conducted in this work argues in favor of the proposed concept, and demonstrates that part of the external anal sphincter innervation comes from pudendal nerves and that the pudendal nerve compression in the pudendal canal may be the cause of pudendal neuralgia.

CONCLUSION

Our anatomical study confirms the existence of separate innervations of the anterior and posterior parts of the external anal sphincter muscle as well as for the pubococcygeus muscle. It may be possible to better identify the site of compression of the pudendal nerve by separate electrophysiological examinations of the three zones. The concept of staged sacral reflexes is introduced and should lead to apply more precisely infiltration treatments or to start surgical interventions.

ABBREVIATIONS

LAN: levator ani nerve; PNA: pudendal nerve afferents; PNE: pudendal nerve efferents, MT: medullair transfer; SC: spinal cord.

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