Original article

Pelvic trauma and pudendal syndrome (post-traumatic pudendal syndrome)

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Abstract: The pudendal syndrome can result from pelvic trauma but the link is difficult to prove. The study was intended to demonstrate the existence of post-traumatic pudendal syndromes. Two case reports were used to illustrate the point. The study was based on 394 female patients of whom 216 (54.8%) had a history of pelvic trauma. Patients were further classified into mutually exclusive groups according to presence/absence of perineodynia and latency of pain appearance in those with pelvic trauma (pain before trauma, early onset, or late onset). Urge urinary incontinence, cystalgia, anal incontinence and proctalgia fugax were found statistically more frequent in the pelvic trauma group. Perineodynia visual analog score, NHI-CPSI score and Wexner's score were also significantly greater in traumatic patients. The three pudendal syndrome clinical signs were significantly more present in pelvic trauma patients than in non-trauma subjects. In the three post-traumatic perineodynia groups with different latency, only minor significant symptom frequency differences were observed but importantly urge incontinence, cystalgia, anal incontinence and proctalgia fugax remained more frequent than in the non-trauma perineodynia group for similar pain scores. Post-traumatic pudendal syndrome is a reality. Perineodynia, urge incontinence, anal incontinence, proctalgia fugax and cystalgia are the most frequently symptoms encountered. These findings recommend performing a detailed history search for any symptom of the pudendal syndrome and a comprehensive clinical examination including its three clinical signs after any significant pelvic trauma.

Keywords: Bladder pain; Incontinence; Medico-legal; Pelvic trauma; Pudendal neuralgia.

INTRODUCTION

Pudendal nerves are involved in the three basic functions of the perineum: defecation, voiding and sexual activity. Because they contain sensory, motor and autonomic fibers, any lesion of these nerves can induce a great variety of symptoms depending on which of the three branches (rectal, perineal, penile/clitoral) and which fibers are compressed or stretched[1-9]. The symptoms vary widely among subjects but in the same patient they are often related to activities affecting the nerves and may be viewed as part of a genuine "pudendal syndrome".

The pudendal syndrome can result from a pelvic trauma but as it is rarely diagnosed patients facing the syndrome after a work or a motor vehicle accident (MVA) have great difficulties proving the link between the trauma and the sequelae that may persist sometimes for the rest of their life.

In the experience of the first author (JB), a history of pelvic trauma (e.g. fall on the tailbone, traffic accident) with or without fracture was found in 60% of the patients who needed a pudendal nerve decompression surgery regardless of the time elapsed between the accident and the appearance of the pudendal syndrome. Amazingly, in the same population, only 5 % of the patients described pelvic trauma as the cause of their symptoms. In a recent study establishing normative values for skin temperature and thermal sensory thresholds in the pudendal nerve territory in a population of presumably healthy women, a history of pelvic trauma was evidenced in only 24.4% of them[10] .

The primary goal of this study was to assess the potential link between pelvic trauma and appearance of a pudendal syndrome. Secondary objectives were to evaluate more carefully perineodynia (perineal pain), one of the salient symptoms of the pudendal syndrome, and its relationship with trauma. Two case reports were used to illustrate the point.

MATERIAL AND METHODS

A retrospective study was carried out on female patients of the perineology [11-14] consultation database of the first author (JB) based on the question: "In your life did you experience any significant trauma that could have had an impact on your pelvic bones (e.g. fall on the buttock-tailbone, professional and/or traffic accident, leg fracture)". A complete history including urinary, ano-rectal and sexual functions was recorded from each patient. Four scores were used to evaluate the main symptoms : ICIQ-SF for urinary incontinence [15], St Mark's score for anal incontinence[16], Wexner's score for constipation [17] and NHI-CPSI adapted for women by Antolak for pain, lower urinary tract symptoms and quality of life[7, 18]. Perineodynia (perineal pain) intensity was measured using a visual analog scale (VAS) from 0 (no pain) to 10 (extreme pain). Three clinical signs characteristic of the pudendal syndrome were checked (asymmetric pinprick sensibility of the perineum, painful skin rolling test and painful pudendal nerve during vaginal or rectal examination)[8]. Perineal descent (and perineal position at rest and during Valsalva's maneuver) was measured with a perineocaliper® [19]. Lack of any score value, clinical sign evaluation or information about urinary incontinence, urinary frequency, nocturia, dysuria, cystalgia, dyspareunia, sexual arousal syndrome, anal incontinence (gas, liquid and solid), dyschesia, proctalgia fugax and perineodynia was defined as an exclusion criterion.

Pelvic trauma and pudendal syndrome

The study material consisted of 394 female patients; 216 (54.8%) had a history of pelvic trauma (17 from MVA) and 178 (45.2%) had not. When comparing the two groups of patients with respect to age, weight, height and parity, no significant difference was observed.

Pelvic trauma and perineodynia

The link between perineodynia, one of the main symptoms of the pudendal syndrome, and pelvic trauma was scrutinized in detail. Proctalgia fugax, a short lasting pain in the anus, was not considered as perineodynia and was therefore studied separately.

The study patients were further classified in 6 mutually exclusive groups according to presence or absence of peri-

neodynia, the occurrence or not of a pelvic trauma, and the latency between pain appearance and pelvic trauma. Seven subjects could not be allocated because latency was unknown, leaving only 387 patients for group analyses. Specifically, the 6 groups were defined as follows.

Patients with a VAS pain score equal to 0 ("no pain", n=103) were distributed in 2 groups:
– Group 1: "No pain- No trauma" (n=65)
– Group 2: "No pain-Trauma" (n=38)

Patients with a VAS pain score > 0 ("pain", n= 284) were split into 4 groups:
– Group 3: "Pain-No trauma" (n=113): perineodynia without any history of pelvic trauma.
– Group 4: "Pain early onset-Trauma" (n=35): early onset of post-traumatic perineodynia. Appearance of pain just after trauma (latency trauma-beginning of pain = 0)
– Group 5: "Pain late onset-Trauma" (n=119): late onset of post-traumatic perineodynia. Appearance of pain several months after trauma (latency trauma-beginning of pain > 0)
– Group 6: "Pain before- Trauma" (n=17): pelvic trauma after the appearance of perineodynia (latency trauma-beginning of pain < 0).

Other early onset post-traumatic symptoms were retrieved from the database.

Case Reports

Two examples of pelvic trauma resulting in pudendal syndrome were selected manually from the personal patient database of the second author (SA).

Statistics

Data were summarized as mean and standard deviation (SD) or as median and interquartile range (IQR) for quantitative variables and as numbers and percentages for categorical findings. Mean values were compared by one-way analysis of variance (ANOVA) or by the Kruskal-Wallis non parametric test for skewed data; the comparison of proportions was done by the chi-squared test for contingency tables. Results were considered significant at the 5% critical level (P<0.05). All calculations were done with SAS version 9.4 (SAS Institute, Cary, NC, USA) statistical package and R version 3.2.2.

RESULTS

Pelvic trauma and pudendal syndrome

Patients and pelvic trauma

When comparing patients with pelvic trauma and patients without pelvic trauma, the only significant difference observed was a reduced frequency of grade 2 or 3 cystocele in the first group (Table I). This could be explained by a selection bias inherent to the perineology consultation database which mainly consists of patients with two types of problems: perineodynia and/or prolapse and urinary incontinence. Four common factors inducing the pudendal syndrome, namely perineal descent, parity (number of deliveries), transobturator mesh surgeries (TVT-O®, Prolift®) and cycling, were equally prevalent in the two groups. Pelvic trauma was associated with fracture or luxation in only 11% of the cases.

Pelvic trauma and symptoms of pudendal syndrome

After adjusting for the difference in the distribution of cystocele grade 2 or 3 reported above, urge urinary incontinence, cystalgia (bladder pain), anal incontinence and proctalgia fugax were found statistically more frequent in the pelvic trauma group (Table II). Perineodynia VAS score, NHI-CPSI score and Wexner's score were also significantly greater in patients with trauma. The proportion of patients with perineodynia (VAS score > 0) was significantly higher in the pelvic trauma group than in the other group (82.4% vs.63.5%; P<0.0001).

TABLE I. Characteristics of patients with and without pelvic trauma

Parameter	Without trauma (n=178)	With trauma (n=216)	P-value
Age (years)	50.8 ± 14.2	51.3 ± 13.2	0.72
Parity	1.7 ± 1.3	1.8 ± 1.2	0.75
Weight (kg)	62.8 ± 11.5	62.7 ± 11.6	0.92
Height (cm)	163.3 ± 6.1	163.7 ± 7.3	0.62
BMI (kg/m²)	23.5 ± 4.2	23.6 ± 4.4	0.88
Biking (%)	19.8	20.9	0.84
Trans-obturator mesh surgeries (%)	4.5	6.5	0.39
Cystocele (%)[a]	21.1	11.8	0.014
Rectocele (%)[a]	12.9	11.3	0.64
Perineum at rest (cm)	0.23 ± 1.0	0.38 ± 1.0	0.18
Perineum during valsalva (cm)	-0.35 ± 1.2	-0.15 ± 1.3	0.14
Perineal descent (cm)	0.58 ± 0.7	0.53 ± 0.6	0.49

[a] grade 2 and 3

TABLE II. Symptoms of the pudendal syndrome in patients with and without pelvic trauma

Parameter	Without trauma (n=178)	With trauma (n=216)	P-value
Stress urinary incontinence (%)[a]	38.8	39.4	0.96
Urge urinary incontinence (%)[a]	21.9	32.9	0.020
Urinary frequency (min)[a]	135 ± 63.8	126 ± 63.7	0.12
Nocturia[a]	0.98 ± 1.3	1.0 ± 1.3	0.68
Dysuria (%)[a]	33.1	40.7	0.063
Cystalgia (%)[a]	22.5	32.4	0.031
ICI-QSF (/24)[a]	4.4 ± 5.1	5.4 ± 6.0	0.26
Perineodynia (/10)	3.7 ± 3.3	4.8 ± 2.9	0.0002
NHI-CPSI (/44)	24.6 ± 9.3	27.8 ± 8.1	0.017
Dyspareunia (%)	39.9	46.8	0.17
Sexual arousal syndrome (%)	20.2	25.9	0.18
Anal incontinence (%)	31.5	41.9	0.034
St Marks score (/24)	3.5 ± 4.7	4.4 ± 5.1	0.22
Prolapse disturbance (%)	20.9	13.6	0.058
Proctalgia fugax (%)	20.2	34.7	0.0015
Dyschesia (%)	75.6	74.5	0.85
Wexner score (/30)	7.4 ± 4.8	9.4 ± 5.6	0.013

[a] P-value adjusted for cystocele

Clinical signs of the pudendal syndrome

The three pudendal syndrome clinical signs were significantly more present in pelvic trauma patients than in non-trauma subjects (Table III)

Pelvic trauma and perineodynia

Pain characteristics

The duration of pain was significantly different in the 4 "Pain" groups (P=0.0002). The median (IQR) pain duration was respectively 22 (10-48) months for "Pain-No Trauma",

TABLE III. Clinical signs of the pudendal syndrome in patients with and without pelvic trauma

Parameter	Without trauma (n=178)	With trauma (n=216)	P-value
Asymmetric pinprick sensibility (%)	40.4	64.4	<0.0001
Painful pudendal nerve (%)	61.2	81.9	<0.0001
Painful skin rolling test (%)	51.7	66.2	0.0035
3 clinical signs positive (%)	28.1	49.1	<0.0001
3 clinical signs negative (%)	27.5	10.6	<0.0001
Number of clinical signs positive	1.5 ± 1.2	2.1 ± 1.0	<0.0001

30 (12-84) months for "Pain early onset-Trauma", 15 (7-48) months for "Pain late onset-Trauma", and 60 (48-96) months for "Pain before-Trauma". The median (IQR) latency between trauma and pain was 0 (0-0) month in the "Pain early onset-Trauma" group and 156 (58-309) months in the "Pain late onset-Trauma" group, while it amounted -36 (-48 to -18) months in the "Pain before-Trauma" group.

Trigger factors

Pelvic trauma was the trigger factor that precipitated symptoms in all 35 patients of the "Pain early onset-Trauma" group. This represents 12.5 % of the patients with perineodynia (20.7% of the patients with perineodynia and a history of pelvic trauma). For the other groups ("Pain-No trauma", "Pain late onset- Trauma" and Pain before-Trauma"), no difference was observed with respect to the trigger factors (Table IV).

TABLE IV. Trigger factors in the "Pain" groups ("Pain early onset-Trauma" excluded)

Trigger factor	Pain-No trauma (N=113) Number (%)	Pain late onset-Trauma (N=119) Number (%)	Pain before-Trauma (N=17) Number (%)	All pain groups (N=249) Number (%)
None	51 (47.7)	55 (50.0)	5 (31.3)	111 (47.6)
Invasive medical procedures or surgeries	16 (14.9)	18 (16.4)	5 (31.3)	39 (16.7)
Long time sitting (work, journey...)	8 (7.5)	8 (7.3)	0 (0.0)	16 (6.9)
Psychological trauma	5 (4.7)	7 (6.4)	2 (12.5)	14 (6)
Wrong movement	7 (6.5)	5 (4.6)	2 (12.5)	14 (6)
Childbirth	7 (6.5)	5 (4.6)	1 (6.3)	13 (5.6)
Cystitis	4 (3.7)	4 (3.6)	0 (0.0)	8 (3.4)
Biking	5 (4.7)	3 (2.7)	0 (0.0)	8 (3.4)
Sexual intercourse	3 (2.8)	2 (1.8)	1 (6.3)	6 (2.6)
Severe dyschesia	1 (0.9)	3 (2.7)	0 (0.0)	4 (1.7)

TABLE V. Symptoms and clinical signs of patients classified in 6 distinct groups as defined in text (N=387)

Parameter	Group 1 No pain - No trauma (n=65)	Group 2 No pain - Trauma (n=38)	Group 3 Pain - No trauma (n=113)	Group 4 early onset-trauma (n=35)	Group 5 pain late onset-trauma (n=119)	Group 6 Pain before-trauma (n=17)	P-value** Group 1 vs Group 2	P-value** Group 3 vs Groups 4-6
General characteristics								
Parity	2.0 ± 1.2	2.3 ± 1.0	1.5 ± 1.3	1.3 ± 1.1	1.7 ± 1.3	2.0 ± 1.4	0.076	0.19
Cystocele grade 2 et 3 (%)	40.3	23.7	10.1	2.9	13.0	0.0	0.040	0.59
Rectocele grade 2 et 3 (%)	19.4	21.1	9.2	11.4	9.5	6.2	0.95	0.91
Symptoms								
Stress urinary incontinence (%)*	24.6	39.5	5.3	8.6	10.1	5.9	0.72	0.22
Urge incontinence (%)	36.9	65.8	13.3	34.3	20.2	47.1	0.020	0.012
Urinary frequency (min)	128.1 ± 63.2	99.1 ± 51.2	139.5 ± 64.0	122.6 ± 61.4	134.4 ± 67.5	144.7 ± 52.2	0.028	0.46
Nocturia	1.1 ± 1.2	1.2 ± 1.8	0.9 ± 1.3	0.6 ± 0.8	1.0 ± 1.2	1.1 ± 1.2	0.90	0.71
Dysuria (%)	40.0	31.6	29.2	31.4	47.1	47.1	0.49	0.0071
Cystalgia (%)	18.5	15.8	24.8	34.3	34.4	41.2	0.62	0.049
ICIQ-SF (/24)	6.2 ± 6.3	11.9 ± 4.3	3.7 ± 4.4	3.3 ± 5.5	4.6 ± 5.8	4.9 ± 4.9	0.070	0.72
Perineodynia (/10)	0.0 ± 0.0	0.0 ± 0.0	5.8 ± 2.2	6.1 ± 1.9	5.6 ± 2.0	7.2 ± 1.8	NA	0.56
NHI-CPSI (/44)	18.9 ± 9.9	20.1 ± 8.0	27.0 ± 7.9	26.7 ± 7.8	29.3 ± 7.1	32.8 ± 6.0	0.41	0.062
Dyspareunia (%)	29.2	28.9	46.0	57.1	47.9	52.9	0.64	0.68
Sexual arousal syndrome (%)	10.8	18.4	25.7	28.6	26.9	23.5	0.22	0.47
Anal incontinence (%)	35.4	34.2	29.2	42.9	43.2	41.2	0.81	0.036
St Marks score (/24)	3.5 ± 5.2	3.8 ± 5.6	3.5 ± 4.6	2.9 ± 4.1	5.0 ± 5.3	3.8 ± 3.1	0.28	0.39
Prolapse disturbance (%)	37.5	29.7	11.1	9.4	9.6	17.6	0.91	0.95
Proctalgia fugax (%)	10.8	18.4	25.7	42.9	36.1	41.2	0.41	0.014
Dyschesia (%)	88.9	38.1	69.8	86.4	76.6	100	0.0020	0.15
Wexner score (/30)	6.0 ± 4.1	5.2 ± 3.2	8.0 ± 4.9	9.7 ± 5.3	10.1 ± 5.5	10.8 ± 7.6	0.16	0.035
Clinical signs								
Asymmetric Pinprick sensibility (%)	29.2	60.5	46.9	62.9	66.4	58.8	0.0008	0.0030
Painful Alcock's canal (%)	35.4	57.9	76.1	97.1	85.7	82.4	0.014	0.027
Painful skin rolling test (%)	36.9	39.5	60.2	85.7	65.6	88.2	0.70	0.094
3 signs negative (%)	47.7	26.3	15.9	2.9	7.6	11.8	0.023	0.030
3 signs positive (%)	15.4	31.6	35.4	57.1	50.4	58.8	0.027	0.0069
Number of clinical signs	1.0 ± 1.1	1.6 ± 1.2	1.8 ± 1.1	2.5 ± 0.7	2.2 ± 1.0	2.3 ± 1.0	0.0093	0.0027

* moderate or severe

** P-values adjusted for grade 2-3 cystocele, rectocele and parity

Relation between trauma, pain occurrence and onset (6 study groups)

Parity, cystocele and rectocele grade 2 or 3 were more frequent in the two "No pain" groups (Table V). The other characteristics were comparable in the 6 groups. Results for symptoms and clinical signs are presented in Table V. P-values were adjusted for parity and grade 2 and 3 cystoceles and rectoceles. Comparisons were made between groups with P-values being adjusted for parity, grade 2 and 3 cystoceles and rectoceles.

Impact of trauma (with or without pain)

« No pain » groups (group 2 versus group 1): In the "Trauma" group (No pain-Trauma, group 2), dyschesia was less frequent (38.1 versus 88.9; P=0.0020) than in the "No pain-No trauma" group (group 1). Further, the rate of urge incontinence (65.8 versus 36.9%, P=0.020) was higher and so were the urinary frequency (99.1 ± 51.2 versus 128 ± 63.2 min, P=0.028) and the number of clinical signs (1.6 ± 1.2 versus 1.0 ± 1.1, P=0.0093). The only clinical sign whose prevalence was comparable in the two groups (with or without trauma) was "painful skin rolling test" (39.5 versus 36.9%, P=0.70).

« Pain » groups (groups 4-5-6 versus group 3): In the "Trauma" groups (groups 4-5-6), urge incontinence (25.7 versus 13.3%, P= 0.012), dysuria (43.9 versus 29.2%, P=0.0071), cystalgia (35.1 versus 24.8%, P=0.049), anal incontinence (42.9 versus 29.2%, P=0.036), proctalgia fugax (38 versus 25.7%, P=0.014) were more frequent than in the "Pain-No trauma" group (group 3). Likewise, Wexner's score values (10.1 ± 5.7 versus 8.0 ± 4.9, P=0.035) and the number of clinical signs were higher (2.3 ± 0.9 versus 1.8 ± 1.1, P =0.0027). The only clinical sign equally prevalent in the two groups (with or without trauma) was "painful skin rolling test" (71.9 versus 60.2%, P=0.094).

Differences between post-traumatic perineodynia groups

"Pain early onset-Trauma "versus "Pain late onset-Trauma": The only significant differences were a lower number of night time voids (nocturia, 0.60 ± 0.81 versus 1.05 ± 1.20, P=0.039) and a greater frequency of "painful skin rolling test" in the "Pain early onset-Trauma" group (85.7% versus 65.5%, P=0.022).

"Pain early onset-Trauma" versus "Pain before-Trauma": The NHI-CPSI score was greater in the "Pain before-Trauma" group (32.8 ± 6.0 versus 26.7 ± 7.8, P=0.037), while perineodynia VAS score tended to be higher (7.2 ± 1.8 versus 6.1 ± 1.9, P=0.055).

"Pain late onset-Trauma" versus "Pain before-Trauma": Urge incontinence (47.1 versus 20.2%, P=0.014) was more frequent and the perineodynia VAS score was greater (7.24 ± 1.79 versus 5.56 ± 1.99, P=0.0014) in the *"Pain before-Trauma"* than in the late onset group but for dyschesia (100% versus 76.6 %, P=0.051) only a tendency was observed.

Other early onset post-traumatic symptoms

Because post-traumatic symptom latencies were not systematically recorded, they could not be analyzed. For urinary symptoms, latencies between trauma and appearance of the problem were available but not specifically for each symptom. The group called lower urinary tract disorders (LUTD) included urinary incontinences (stress and urge), urinary frequency, urgency and dysuria. Of the 95 latency cases available, LUTD appeared just after pelvic trauma in 9 cases, 8 of which together with perineodynia ("Pain early onset-Trauma").

Latencies were also available for sexual troubles as a group (dyspareunia and sexual arousal syndrome). In 3 cases out of 50 available, sexual disorders appeared immediately after trauma (one of these cases was a sexual arousal syndrome associated with dyspareunia). The 3 cases arose together with perineodynia and LUTD.

Latencies for anal incontinence were available for 75 cases. This symptom appeared directly after trauma in 8 cases. In 2 of them, it was the only early onset symptom. Further, it was either associated with perineodynia, sexual troubles and LUTD (n=2), appeared together with perineodynia and LUTD (n=2) or it arose with perineodynia only (n=2).

Case reports

Patient 1 - A 53 year old female was driving through an intersection when her auto was struck on the driver's side by another auto traveling at a high rate of speed. Her automobile was lifted an estimated six feet in the air, rotated and landed upright. She was wearing a seat belt and air bags deployed. She suffered immediate pains in the left leg, left foot and left buttock. Pelvic fractures were not identified on CT scanner evaluation. Within 10 to 14 days, she noted perineal pain and a sensation of "sitting on one of her sitting bones". She developed urinary frequency and double voiding that had not been present in the past. There was gradual onset of persistent genital arousal. Underclothing began to cause the left labial pain. For several months, treatments focused on her left leg and foot and included a neurotomy of a distal portion of the sural nerve. Progressive increase in pelvic pains, originally ignored by caregivers, resulted in referral for evaluation. There was no history of previous pelvic pain. Since the MVA, she was unable to work as an accountant because sitting was too painful. The persistent pain impaired cognitive function. Pain distracted her thinking, reduced her mental acuity and affected her efficiency.

Sensory examination in the pudendal territory revealed hypoalgesia at the left side of the clitoris and hyperalgesia at the right side of the clitoris and left anal site. Pinprick was normal at the labia and right anal site. Pelvic floor descent was < 1 cm. There was mild erythema of the labia majora. The skin over the coccygeal area and natal cleft was smooth and pale suggesting trophic changes.

In the 16 months prior to diagnosis of pudendal neuropathy unsuccessful treatments had included sacral iliac joint block, caudal epidural steroid injection, sympathetic nerve block. She had slight, transient pain reduction from pelvic floor physical therapy and also when using a transcutaneous nerve stimulator.

Additional neuropathic pain generators were identified by using palpation and skin *rolling* [or "pinch roll"] test (thoracolumbar junction syndrome, ilioinguinal and iliohypogastric nerves bilaterally) [20] .

She had limited relief with a series of three pudendal blocks and was advised to undergo decompression surgery of the pudendal nerve. She is recovering from that operation with early improvement in her pudendal syndrome.

Patient 2 - A 56 year old woman was stopped at a traffic light when her heavy sports utility vehicle was struck from the rear by an auto traveling at approximately 56 km/h. Seat belt was being worn. She had immediate low back pain, a transient "electric shock" pain in the left lower quadrant and leg and a marked sensation of nausea. Neck and shoulder pain developed over a few days. Emergency evaluation included magnetic resonance imaging of the lumbar spine. No acute abnormalities were identified. She

had physical therapy, acupuncture and multiple medications for the low back and leg pains.

Five months after the MVA she began to notice rectal pain while driving her auto. Dyspareunia developed. Over several weeks the rectal pain became severe. She was unable to sit in her job. There were occasional episodes of fecal and urine incontinence. A foreign body sensation developed in the perineum. Pains became progressively worse and required use of oral morphine for control.

Eighteen months after the MVA the diagnosis of pudendal neuropathy was made by second author (SA). Pinprick caused hyperalgesia at the clitoris and labia (right > left). Skin changes of cutis anserina and dilated pores were observed over the coccygeal area. There was no prolapse and perineal descent with Valsalva maneuver was < 1.5cm. No additional pelvic pain generators were found at examination. The pudendal nerve terminal motor latency was prolonged bilaterally, showed temporal dispersion and caused hyperpathia (after sensation). The warm temperature threshold detection test was abnormal[10, 21].

A series of three pudendal nerve blocks was performed. Each relieved her pain but only for seven to ten days. Decompression surgery was recommended but was postponed because of insurance problems. After additional pudendal nerve blocks for pain control the surgery was completed five years after the MVA. Early, complete pain relief lasted only a few weeks. Rectal pains returned. She has not required post-operative pudendal nerve blocks. Central sensitization requires continued medications.

DISCUSSION

Pelvic trauma and pudendal syndrome

Based on case reports and clinical experience, pelvic trauma is a well-known trigger factor for pudendal neuralgia[7, 22]. This study is the first one comparing all symptoms of the pudendal syndrome in female patients with or without trauma of comparable age, parity and biometry.

History of pelvic trauma significantly increased frequency of urge incontinence, perineodynia, cystalgia, anal incontinence and proctalgia fugax. Wexner's score and NHI-CPSI score were also greater in case of trauma. Other symptoms of the pudendal syndrome (urinary frequency, dysuria, sexual arousal syndrome, dyspareunia) were more frequent after trauma but not significantly so because of the characteristics of the studied population (high prevalence of pudendal syndrome). Around 10% of LUTD, sexual troubles and anal incontinences began just after pelvic trauma.

Because the pudendal syndrome can be the consequence of a pelvic trauma and because it is rarely diagnosed, patients facing this problem after a professional or traffic accident have great difficulties to prove the link between trauma and sequelae that might persist for the rest of their life. In an attempt to explain the possible causal effect of pelvic trauma on pudendal syndrome, the expert could be guided by the seven Simonin's criteria which can be adapted for post-traumatic pudendal syndrome (these criteria are similar to those used in the English and American legal and medical systems to develop evidence of causation for both industrial problems and medical issues) [23-27]. Specifically, these criteria are:

C1. *The nature and intensity of the trauma*: pelvic trauma must be clearly documented and enough important to explain the lesion.

C2. *The quality and quantity of symptomatology*: the link between pelvic trauma and pudendal syndrome must be clinically possible and logical.

C3. *The coherence of impact-anatomical site-and type of injury su*stained: directly or closely related.

C4. *The natural history of the condition*: the symptoms have to follow a logic evolution since the trauma.

C5. *The elapsed time between event and complaint*: latency between trauma and pudendal syndrome must agree with the clinical experience.

C6. *The pre-morbid state of the claimant*: the pudendal syndrome didn't exist before the trauma.

C7. *Exclusion of another etiology of pudendal syndrome*: like a delivery, a trans-obturator surgery, biking or any other.

Regarding the importance of the trauma which could induce a pudendal syndrome (Criterion 1: Nature and intensity of the trauma), it is important to note that 89% of the trauma included in this study were not associated with a luxation or a fracture. It is therefore not necessary to have an organic – radiological lesion to have severe perineal sequelae.

Functional and/or organic damages to the pudendal nerves (and the other nerves involved in perineal innervation) after a pelvic trauma is easily understandable (Criterion 2) if one compares the pelvic nerves with electric cables in a car chassis (pelvic bones, fascia and muscles). Pudendal nerves have a tortuous and narrow path going between the anterior part of the piriformis muscles and the posterior border of the sacro-spinal ligament. Then passing through the interligamentary space between the sacrospinous (and its muscular part, the coccygeal muscle) and sacro-tuberous ligaments, through the fascia lunata and, at last enclosed in the Alcock's canal lying on the obturator muscle. Furthermore, some branches of the pudendal nerve may pass directly through the ligaments. Each trauma can create a distortion of the fibrous part (ligaments and fascia) of the pudendal nerve path. Because the chassis has moved, the electric cables are no longer in the same position and electrical (neuritic) short circuit can occur. Reflex muscle contractures, trigger points (piriformis, obturator, coccygeus, levator ani) directly induced by the trauma or secondary to neuralgias (the muscles tried to avoid any nerve movement), abnormal posture or psychological distress could be responsible for more nerve compression or stretch. In such a case, the nerve begins to swell and cannot move any more freely in its path giving rise to more pain. The vicious cycle is ongoing. Furthermore, pelvic hematoma or callus formation around a fracture may cause perineural scarring and compression. Results presented in Table II and Table III clearly show that the link between pelvic trauma and pudendal syndrome is clinically possible and logical (Criterion 2).

If the patient experienced a significant trauma that could have an impact on her pelvic bones (fall on the buttock-tailbone, traffic accident, leg fracture…), there is a coherence of impact-anatomical site-and type of injury sustained (Criterion 3).

For Criterion 4, the symptoms have to follow a logic evolution since the occurrence of the trauma (natural history of the condition). Of course, there is no problem in case of early onset post-traumatic symptoms. But, to fulfill this criterion for late onset post-traumatic symptoms, a prospective study should be considered to describe precisely the natural history of the condition (e.g. latency between pelvic trauma and each symptom occurrence, spontaneous evolution of the syndrome with time…).

Criterion 5 corresponds to the elapsed time between pelvic trauma and appearance of the pudendal syndrome. It is probably the most important one. This aspect has been studied carefully for perineodynia, one of the key symp-

toms of pudendal syndrome, because latencies were available for almost all the patients.

Pelvic trauma and perineodynia

Normally, pain is triggered by the stimulation of pain receptors (nociceptive pain) but in neuralgia (neuropathic pain) it occurs without excitation of these receptors and is caused by an abnormal change in the structure or function of the nerves. Innervation of the perineal skin is mainly provided by the pudendal nerves but other nerves are slightly involved (ilio-inguinal, genito-femoral, perineal branch of the posterior femoral cutaneous nerve, inferior cluneal).

Pudendal neuralgia is usually defined according to the Nantes criteria. These criteria are quite weak and they were never validated clinically. In fact, three of the five essential criteria are unreliable.

The first key criterion is an increase of pain while sitting. According to the Nantes team itself the same arises in case of inferior cluneal neuralgia but on harder seats [28] indicating that this sign is not specific.

The second one - Pain with no objective sensory impairment – does not reflect clinical reality. In fact, in Shafik's studies[5, 6] and in our personal experience[7, 8], pinprick hypoesthesia and hyperesthesia is often found in case of perineodynia. This is logical because nerve damage can be responsible for numbness or allodynia like in other parts of the body. Increase of warm detection threshold (reduced sensitivity to warmth) and qualitative abnormalities during this test (habituation, allodynia, dysesthesia, radiation or aftersensation) are also frequently encountered in case of pudendal (and other) nerve damage [21, 29].

A positive pudendal block (clear reduction of pain during anesthesia time) is a third weak criterion. According to the Nantes team itself: "a positive pudendal block is not specific as it simply indicates that the pain is situated in the territory of the pudendal nerve; pain related to any perineal disease (e.g., anal) would also be relieved by pudendal nerve block. A negative block does not formally exclude the diagnosis when it is not performed with sufficient precision"[30]. Therefore according to the Nantes team, this test is neither specific nor sensitive. It is even less reliable if perineal sensation is not checked 2 hours after the block [31].

The term perineodynia[8], a clear symptom, should be recommended rather than pudendal neuralgia, only one of the possible etiologies, because its definition criteria are weak and other nerves can be involved in perineal pain.

What about the cause and effect relationship in the 4 groups with a history of trauma?

The easiest situation is the early onset perineodynia ("Pain early onset-Trauma" group) in which pain arises just after pelvic trauma. In such a case, the cause – effect relationship could be evident (Criteria 1 to 5 positives) if there is no pre-existing pudendal syndrome in the medical history of the patient (Criterion 6) and if there is no other trigger factor (Criterion 7). Painful symptoms are frequently associated with perineodynia (cystalgia, dyspareunia and proctalgia fugax) together with urge incontinence, anal incontinence and dyschesia.

The medico-legal evaluation is the same in case of early onset post-traumatic LUTD, sexual troubles or anal incontinence without pain. This situation corresponds to patients in the "No pain-Trauma" group. More difficult are the "Pain late onset-Trauma" and "Pain before-Trauma" groups. In the "Pain before-Trauma" group, perineodynia means a problem already exists on the pudendal nerves (or other nerves involved in perineal sensibility) before pelvic trauma. Trauma just aggravates the pre-existing pudendal syndrome. The medico-legal discussion will be about a possible worsening of the syndrome after trauma. Our study suggest that it should be the case because the "Pain before-Trauma" group is notably different from the "Pain-No trauma" group. To conclude at a clear cause-effect relationship, the patient should ideally have an evaluation of the pre-existing pudendal syndrome to search for a clinical worsening.

In case of "Pain late onset-Trauma", it seems that the pudendal syndrome with trauma is worse than without trauma (with the same triggering factors). It is like if the trauma created a borderline state waiting for a trigger factor to develop more severe symptoms. Probably other symptoms of the pudendal syndrome already existed before pain appeared like in the "No Pain-Trauma" group which is possibly a step before "Pain late onset-trauma". In a medico-legal situation it is mandatory to ask about symptoms pre-existing before pain (especially all types of lower urinary tract dysfunctions, sexual arousal syndrome, anal incontinence and proctalgia fugax).

The 3 clinical signs of pudendal neuropathy-pudendal syndrome seem to be correlated with the severity of the syndrome. The skin rolling test is more frequently painful in case of perineodynia probably because it is explained by an allodynia to skin pinching. Pinprick sensibility is more frequently asymmetric in case of pelvic trauma maybe because it reflects a nerve compression or stretch.

Case reports

Case 1 - This woman did not notice immediate perineal pain. Its onset was minimally delayed (10-14 days) and should be considered as a "Pain early onset-Trauma" case. LUTS slowly developed. Persistent genital arousal appeared later. The time elapsed before diagnosis and treatment of the pudendal syndrome was 16 months which is quite typical. A correct diagnosis just after the MVA would have allowed avoiding useless treatments and unnecessary psychological and physical suffering. Furthermore, because the MVA was induced by another driver, the pudendal syndrome was part of the personal injury. This was completely ignored until treatment of the pudendal neuropathy was going on. Medico-legal actions were pursued however the long latency before a correct diagnosis caused significant medical expenses for the leg injury and loss of income.

Case 2 - Onset of perineal (rectal) pain, incontinence, and dyspareunia was delayed for several months ("Pain late onset-Trauma"). The diagnosis of pudendal neuropathy was not made for almost three years. In the meantime, treatment focused initially on back and leg pains. Legal action had started before the diagnosis of the pudendal syndrome. Delayed diagnosis of pudendal neuropathy caused loss of employment, personal stress and the expense of many medical interventions.

Study limitations and future prospects

This retrospective study has some limitations. The study population was characterized by a high prevalence of pudendal syndromes, prolapses and incontinences. Because of the retrospective aspect of the study, data concerning the type or even the existence of trauma and the timing of the symptoms was highly dependent on the patient's memory and data collection during the past consultations. Further, only the latency between trauma and perineodynia was available for analysis. Too many values of latency were lacking for lower urinary tracts, sexual and ano-rectal symptoms to make a comprehensive analysis and to draw firm conclusions.

A prospective study would be ideal. In such a study, after each pelvic trauma, clinical signs and symptoms (and questionnaires) of pudendal syndrome should be sought carefully and the follow-up duration should be at least 1 year. The patient should be warned to contact immediately the physician in case of any new perineal symptom. Latency between each symptom and the trauma should be listed to precisely characterize the classical evolution (probably different types) of the post-traumatic pudendal syndrome (Criterion 4).

Post-traumatic management of male patients should follow the same path. Particular attention is required to the appearance of impotence (partial or complete), ejaculatory impairment (loss of pleasure, pain), "prostatitis-like" pain and sexual arousal syndrome[3, 7, 32].

Faced with a medico-legal situation it is mandatory to be even more precise and in case of doubt to utilize a warm and cold detection test, a pudendal nerve terminal motor latency test and a complete electromyographic (EMG) exploration. Of course, characteristics of the trauma must be defined as precisely as possible (Criterion 1). The patient must be followed up carefully during at least 1 year and advised to inform her doctor in case of any new post-traumatic symptom.

CONCLUSION

According to this study, post-traumatic pudendal syndrome is a reality. Perineodynia, urge incontinence, anal incontinence, proctalgia fugax and cystalgia are the most frequently symptoms encountered. A complete history searching for any symptom of the pudendal syndrome and a clinical examination including its three clinical signs should be done after any significant pelvic trauma. In medico-legal situations it is suggested to add a warm and cold detection test and an EMG evaluation with latencies as soon as possible and one 1 year later in addition to the clinical control.

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