Neurosurgical treatment of perineal neuralgias

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Abstract

Perineal pain is the basis of presentation to different specialities. This pain is still rather unknown and leads the different teams to inappropriate treatments which may fail.

For more than twenty years, we have seen these patients in a multidisciplinary consultation. Our anatomical works have provided a detailed knowledge of the nervous supply of the perineum which allowed us to propose the description of an entrapment syndrome of the pudendal nerve. Other disturbances of different origins were highlighted helping colleagues to a better analysis of this enigmatic painful syndrome.

Cadaveric studies have been done to guide treatments by blocks and surgery if necessary according to well defined criteria.

A randomized prospective study validated the surgery. The retrospective study concluded that two thirds of the patients improved after treatment. New anatomical concepts are leading us to enlarge the field of this type of surgery, with the hope of improving the success rate.

Keywords: Perineal pain; pudendal neuralgias; Alcock's tunnel syndrome; pudendal nerve surgery; blocks of the pudendal nerve.

Introduction

Perineal neuralgias are usually misunderstood and lead patients to medical nomadism and misdiagnosis.

Very often specialised medical teams relate such pain to the pathology of one organ, more often pelvic than perineal. The treatment is then guided by this target organ and leads to surgical techniques (hysterectomy, prostatectomy . . .) which fail and leave a disappointed patient. Radiological data is verified to be normal and then we may think that the pain arises from a truncal palsy. The fact is that the pain is situated in the sensory territory of the pudendal nerve and that it occurs when in the seated position. We undertook an anatomical study to define possible sources of entrapment along the course of the nerve by positioning cadavers in the seated position. Subsequently a neurophysiological approach led us to consider that in many cases prolonged distal motor latency was usually found just like in carpal tunnel syndrome. Treatment is based on nerve blocks guided by CT scan and surgery by a transgluteal approach. We consider that perineal neuralgia is not commonly a pudendal nerve palsy, and is, an entrapment syndrome in the deep spaces of the buttock.

We will exclude all pain with a local explanation (dermatological infections, tumours and so on).

Anatomy, pathology and nosology

Pelvi-perineal pains (PPP) are not rare. It is estimated that up to 3.8% of women aged between 15 and 73 years old suffer from PPP which is more than migraine (2.1%), identical to asthma, and a little less than lumbar pain (4.1%). 15% of gynaecological consultations are for vulvodynia. Pelvic pain is excluded from this study. The pelvis and the perineal spaces are separated from each other by the levator ani muscle. Local disorders such as tumour, dermatological lesions, haemorrhoid, etc. must be recognized and treated. This chapter considers only perineal neuropathic pain of which three different groups must be distinguished:

Neuropathic somatic perineal pain

The perineum is supplied by somatic nerves which are divided into two groups

- sacral origin: pudendal and inferior cluneal nerves
- thoraco-lumbar origin: ilio-hypogastric, ilio-inguinal, genito-femoral and obturator nerves.

Neuropathic visceral pain

This is mediated by the sympathetic system and constitutes most pelvic pain as the sympathetic system infiltrates all the viscera of this area (interstial cystitis, vulvar vestibulitis, levator ani syndrome, orchialgias ...). Urethralgias are typical perineal visceral pain and may be the equivalent of interstial cystitis. Their presentation is quite different from somatic pain. Nevertheless, sympathetic dysfunction can be noted even when only the pudendal nerve is damaged, as this nerve includes sympathetic fibers.

Myofascial pain

This consists of myofascial syndromes which may be components of fibromyalgia. On the other hand, "single" muscular contractures may lead to pathological disorders. Piriformis or internal obturator muscles syndromes lead to a gluteal pain with an ischiatic component in the inferior limb. They can be encountered with a true pudendal palsy.

Pudendal nerve entrapment

Clinical features [34]

The population is mostly in the 50–70-year old age group, with 60% being females. This incidence may be the result of pregnancy which causes a pulling effect which leads to a kind of fragility of the nerves.

The standard clinical presentation is an adult patient with uni- or bilateral pain in the anal, uro-genital or both areas of the perineum, exacerbated in the seated position [15, 23]. It's a neuropathic pain [7]. Pain is located in the territory of the pudendal nerve. We must be careful and distinguish a scrotal pain (pudendal nerve) from a testicular pain (thoraco-lumbar pathology).

Mostly the onset is insidious, but a lot of patients believe that the starting point of the pain is from a pelvi-perineal surgery, forgetting the fact that this surgery was first done to relieve pain. Anyway, surgery of the hip joint with traction of the leg may induce pudendal nerve compression [11, 32]. Gynaecological surgery with hysterectomy by a trans perineal approach may be responsible for pain as the surgeons hang the vagina with wires around the sacrospinal ligament and may then entrap the nerve in the suture [1, 29, 35].

Sitting on the toilet is not painful. Pain is absent in the early morning and increases during the day, and leads to a complete avoidance of the seated position. Cycling is a classic factor which leads to pain [2]. Driving too. A fall can also be advocated. Standing or lying makes the pain decrease or vanish at the beginning of the symptoms. It is usually a burning pain without any dysfunction of the sphincters and perineal function. Associated signs may lead to a wrong diagnosis: sciatica may be noted, which is incomplete most of time, reaching only the popliteal fossa. The muscular component explains such radiation. Pollakiuria may be noted as well as dysuria, but they are just functional syndromes referred to the pain. Sexual disturbances are not frequent although such activities are avoided because of pain. Constipation is more frequent and leads to perineal efforts which damage the nerve.

Neurological examination is normal (no sensory impairment, no motor weakness, positive reflexes). Intra rectal manipulation reproduces a pain mostly at the ischiatic spine level and in Alcock's canal.

Intractable chronic pain has significant emotional consequences and a psychological component is an integral part of the pain process. After surgery performed on an organ that brings no benefit, patients are in a state of distress being unable to enjoy their lives (no cinema, no restaurant, difficulties for driving cars and bikes or motorbikes, no desire for sex because of the pain).

To summarize:
– it is a neuropathic pain (burning pain) with sometimes allodynia, corresponding to the territory of the pudendal nerve;
– pain increases or only exists in the seated position.

Anatomical datas

We thought from the beginning of our studies that this perineal pain was due to a pudendal nerve pathology. The fact that the seated position would lead to pain led us to believe that an entrapment syndrome might exist [15, 23, 25].
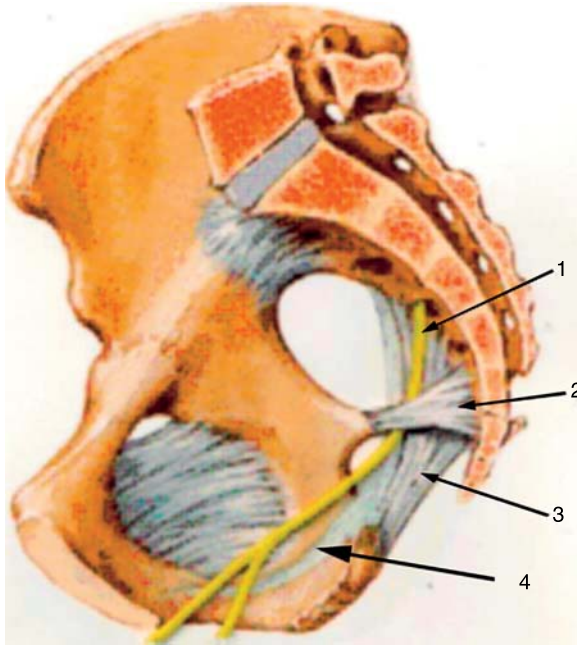
Fig. 1. Medial view of a right hemipelvis showing the course of the pudendal nerve. *1* Pudendal nerve, *2* sacrospinous ligament, *3* sacrotuberal ligament, *4* falciform process

The pudendal nerve (Fig. 1) arises mostly from the S3 root, sometimes from S2 and S4. Its course starts in the presacral area, then it goes laterally to penetrate the greater ischial aperture under the piriformis muscle medial to the ischiatic nerve. In the gluteal region, it crosses behind the distal insertion of the sacro-spinal ligament, then goes medially to enter the pudendal tunnel in the inner aspect of the ischial tuberosity. In its perineal course, the nerve is in the infra-levatori space, and is situated within the internal obturator muscle fascia (Alcock's canal) (Fig. 2). Its branches perforate this fascia to reach their target zones (Fig. 3). The motor branches supply the external striated sphincters of the anus, the urethra and the erector muscles. The sensory fibres supply the skin of the anal region, the intermediate perineum, the penis or the clitoris, the scrotum or the labia.

According to our work on cadavers, three potential entrapment zones can be described:

– In the gluteal region, between the sacro-tuberal and the sacro-spinous ligaments which cross each other;
– At the entry to Alcock's canal by the falciform process of the sacro-tuberal ligament;
– In the pudendal canal, among the splitting of the internal obturator fascia.
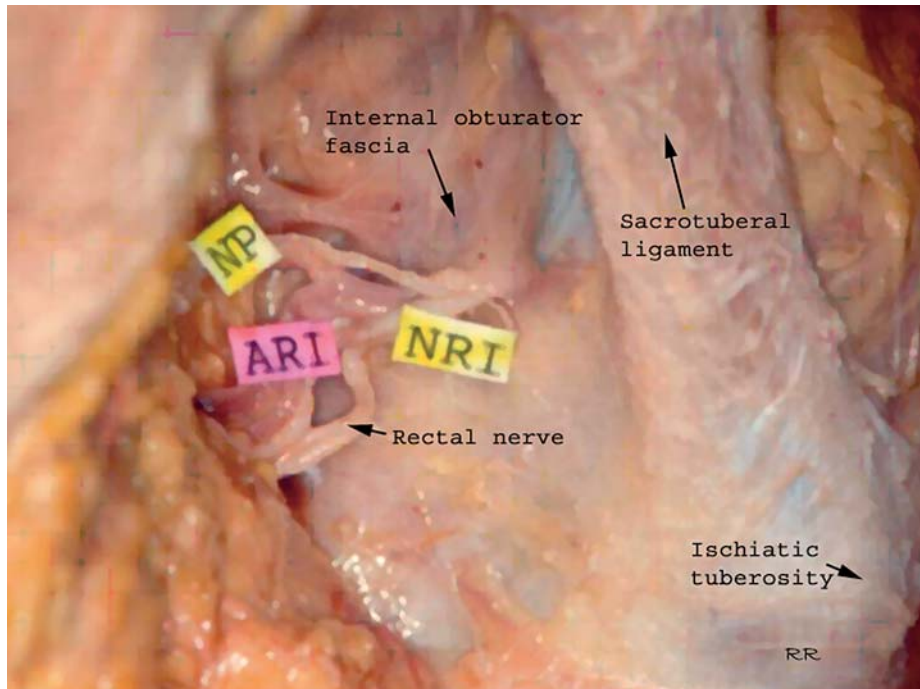
Fig. 2. Medial view of a right hemipelvis. The pudendal nerve starts from mainly the third sacral root in the presacral area, then enters the gluteal region around the sacrospinous ligament, and then goes through the pudendal tunnel in the fascia of the internal obturator muscle which has been resected here

In the seated position on cadavers we can see the ascension of the ischio-rectal fat tissue which leads to compression of the nerve trunk at the 3 levels mentioned above (Fig. 4).

Neurophysiological data

An objective test was required to confirm a compression component. According to evaluation of other well known entrapment syndromes such as carpal tunnel syndrome, we have devised a distal motor latency (DML) test by stimulating with a finger electrode in the rectum, and recording the responses in the erector muscles. This DML must be less than 5 milliseconds [3].

Radiological data

Tumoral pathology must be excluded in the pelvi-perineal area by an MRI or at least a CT scan. Sometimes, calcification of the sacro-spinal ligament or benign osteogenic tumor of the ischiopubic branch can be noted and may induce a compression of the nerve.

Fig. 3. Posterior view of the right side of the entry of the pudendal nerve in the pudendal (Alcock's) tunnel. The proximal branches of the nerve perforate the fascia of the internal obturator muscle to reach the anal region

Treatment

Medical therapy

Medical treatments based on anti-convulsivant and anti-depressant medications for neuropathic pain are insufficient, but can bring mild relief.

Blocks

Such techniques are both diagnostic and therapeutic.

Those techniques must be performed under very standardised conditions [6, 13]. A CT scanner must be used with injection of local anaesthetic and steroids. The patient is in a prone position in the CT scanner. The needle is inserted under local anaesthesia toward the ischiatic spine and displaced in the inner part of it. Paraesthesias may occur which signifies the good position of the injection. Contrast product is then injected, followed by local anaesthetic and steroids. For a block in the internal obturator fascia, the same conditions are required. The needle must be parallel to the muscle, in the inner part of it.
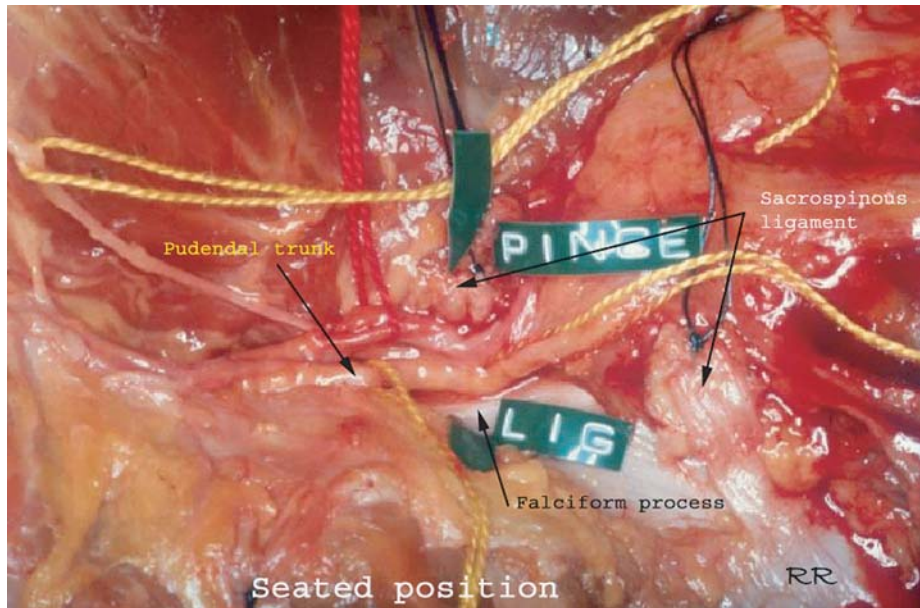
Fig. 4. Medial view of a right hemipelvis. The cadaver is in a seated position. The fat of the ischiorectal fossa lifts and may entrap the nerve in the internal obturator muscle fascia, and in the claw between the sacrotuberal and the sacrospinous ligaments. The falciform process if tall may also damage the nerve

Just after the blocks, the patient is kept in a seated position for half an hour. Then a Visual Analogic Scale (VAS) is established to appreciate the analgesic effect of the local peritruncular block which is an important diagnostic test. The efficacy of the steroid will be judged two or three weeks later. This treatment can be sufficient and done two or three times. Only patients with a positive diagnostic test and no long term side effect of the steroids should be offered surgery.

Surgical procedure

This has been described previously in the literature [23]. We started using a perineal approach in 1986. The problems encountered were deep incision through the firm fat tissue in the ischio-rectal space; problems with hemostasis of huge and numerous veins; difficulties in reaching the main point of entrapment, i.e. the claw between the sacro-tuberal and the sacro-spinal ligaments. There were significant risks of sepsis. Moreover, patients felt pain when sitting on their perineal scar for some days following surgery. Hence, we decided to use a transgluteal approach which has been adopted with good results by other
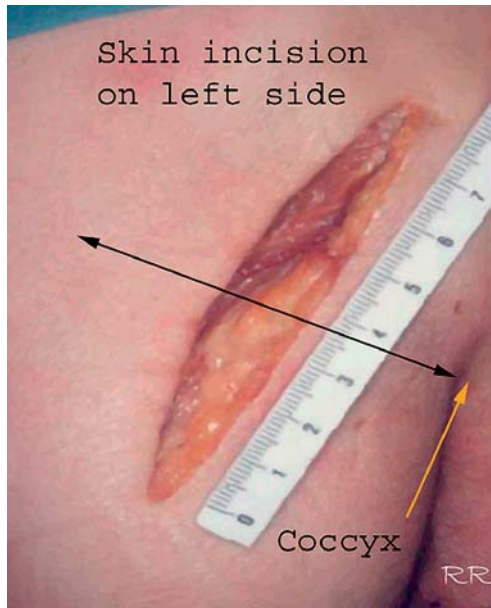
Fig. 5. Skin incision on the left side according to the obliquity of the gluteus maximus muscle fibres

teams [4, 22, 28]. Recently, a transperineal approach has been analyzed nevertheless [5].

Our transgluteal approach [25] allows all possible entrapments that we have detailed to be corrected easily in a single incision. Under general anesthesia, the patient is placed in the so called genu-pectoral position. In this position that we have only been using for a couple of months, the exploration is easier deep in Alcock's canal, better than in the "classical" single lying position. A gluteal incision of about 7 cm in length is made uni or bilaterally on both sides of a transversal line from the top of the coccyx, oriented obliquely according to the direction of the gluteus maximus fibres which are dissected and disinserted from the sacro-tuberal ligament (Fig. 5). The narrow section of this structure is resected transversely with the scissors (Figs. 6 and 7). We must remember that the nerve can go through a layer of the sacro-tuberal ligament and could be severed when cutting it. It is situated at the exact level of the ischiatic spine. The pudendal neurovascular bundle is then visible and released from the dorsal surface of the sacro-spinous ligament. A simple retractor holding medially the ischio-rectal fossa fat is sufficient to open Alcock's canal and to follow the distal branches of the nerve (Fig. 8). If the fascia of the internal obturator muscle is thickened or the falciform process is obstructing, these can be incised. The sacro-spinous ligament is cut and the nerve can be transposed frontally to the ischial spine (Fig. 9).
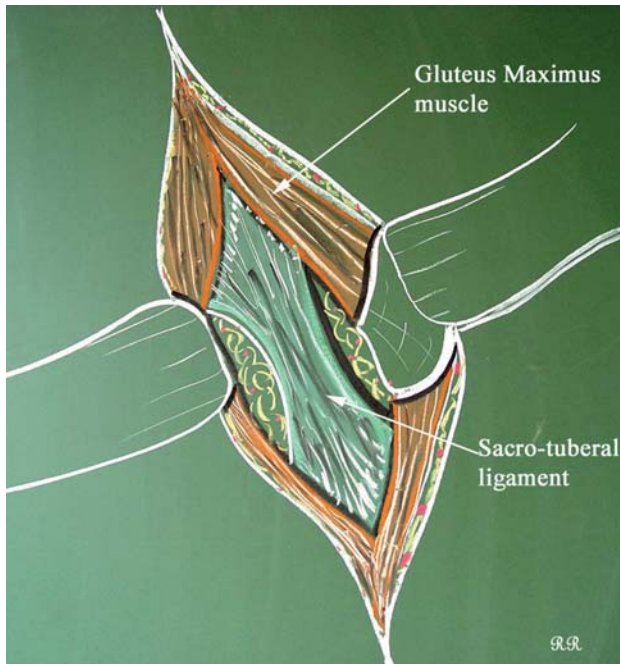
Fig. 6. Surgical approach on the right side of the superficial layers. After the skin incision, the fibers of the gluteus maximus muscle are divided and resected from the posterior aspect of the sacrotuberal ligament

One may then assess the diameter of the nerve and its potential variations [22, 25], its shape (flattened or not), its inflammatory appearance, peritruncular fibrosis and satellite veins dilatations, vasculo-nervous conflict, abnormalities of the nerve course (such as trans sacro-spinous or trans sacro-tuberal pathways) [26] and the shape of the ischiatic spine [4]. The combination of normal anatomical conditions and a normal nerve is a bad prognostic factor of course [26]. The closure is done in two planes. Drainage is usually unnecessary. A 3–5 days hospital stay is required. The duration of this procedure is 20–30 minutes for one side. The sacrifice of the two ligaments has no morbid consequences for the sacro-iliac joint in normal cases. If patients are complaining of a true sacro-iliac instability prior to surgery, we can preserve the sacrotuberal ligament and only cut the sacrospinous one. The approach is then done a little more medially than the standard technique. The nerve is reached in the inner and ventral part of the sacro-tuberal ligament.

Sitting is allowed the day after surgery according to the neuropathic pain. The wound is above the pressure area. All the different pathological considerations during surgery must be clearly noted in a report.
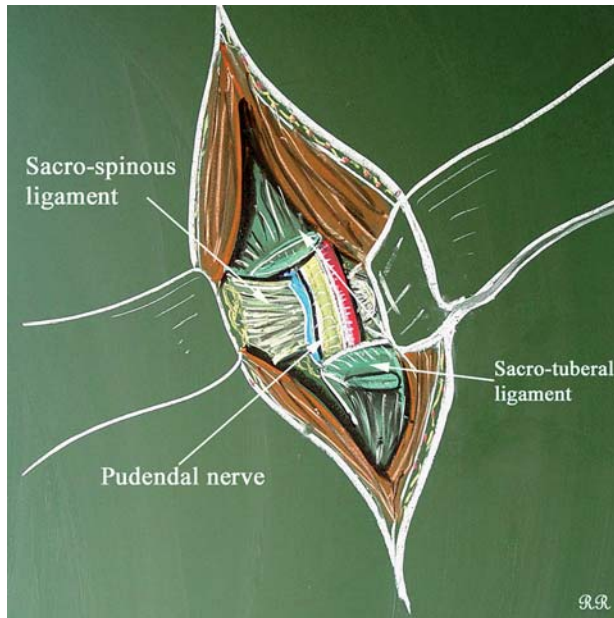
Fig. 7. Surgical approach; the sacrotuberal ligament is cut and resected. The pudendal nerve and vessels appear behind to sacrospinous ligament

Results

We have analyzed our results both in a prospective randomised study and retrospectively.

Randomized prospective controlled trial [27]

Method

Study inclusion criteria: patients eligible for inclusion had chronic perineal pain for at least one year's duration. The pain, uni or bilateral, was exacerbated in the seated position. Patients had to be between the ages of 18 and 70 and had pain intensity of at least 70 on 100 according to the VAS score. We also used a behavioural scale and the Hamilton depression rating scale (minimum score at 3 for the first and 9 or below for the latter).

A positive diagnostic response to an anesthetic block of the pudendal nerve was required and also the persistence of perineal pain in spite of at least two steroid blocks. Every patient had a normal pelvi-perineal CT scan, avoiding all the tumour pathology which could lead to pain.

The study design was a sequential, randomized controlled trial without blinding. Patients were assigned randomly to one of the two treatment groups: surgery or control. The only difference in the treatment between the two
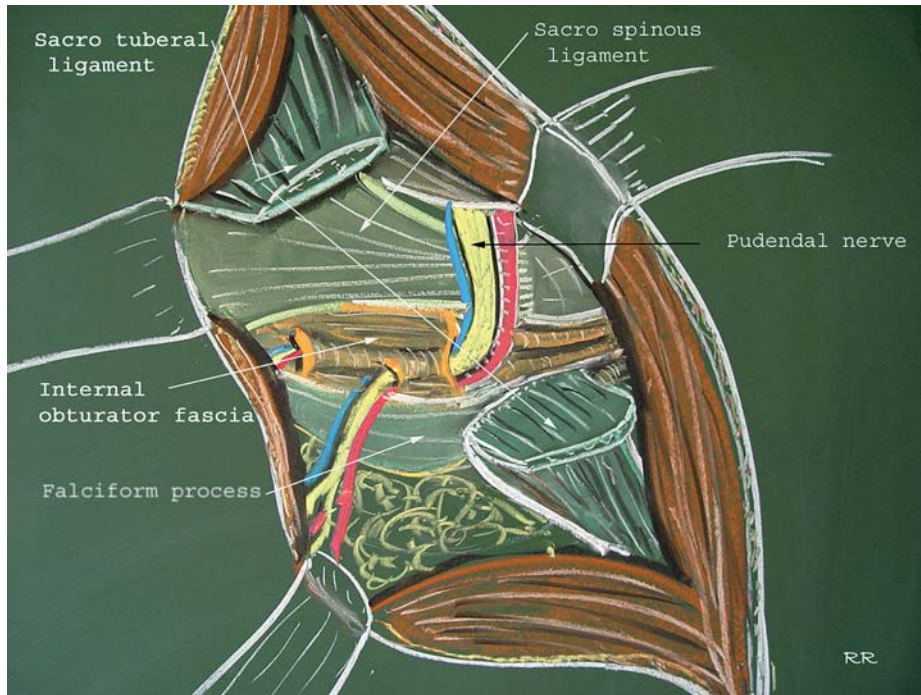
Fig. 8. A deep spreader is used to open the inner part of the ichiatic region. The puden-dal canal appears; the pudendal nerve enters the tunnel in an undoubling of the internal obturator muscle fascia. The anal branch perforates this layer to reach the anal area

groups is related to the fact that only the patients of the first group were operated on.

Outcome measures: the primary endpoint of the study was the proportion of patients judged to have improved three months following surgery or after 3 months of medical treatment. Treatment was considered effective if the pain score had decreased by at least 30 on the VAS and less than 3 on the behaviour scale. We followed up both groups 1 year after the inclusion and the surgical group was assessed again 4 years later to find out whether improvement was maintained.

Results

Thirty-five patients were eligible for the study among 181 patients seen in a multidisciplinary clinic over a two years period. Three patients refused and the other 32 were recruited, 16 to each of the study groups. At 3 months, 50% of the surgery group were improved versus 6% of the control group. At 12 months by analysis to treat protocol, 71% in the surgical group and 13% of the control group had a successful outcome. The surgical procedure is there-
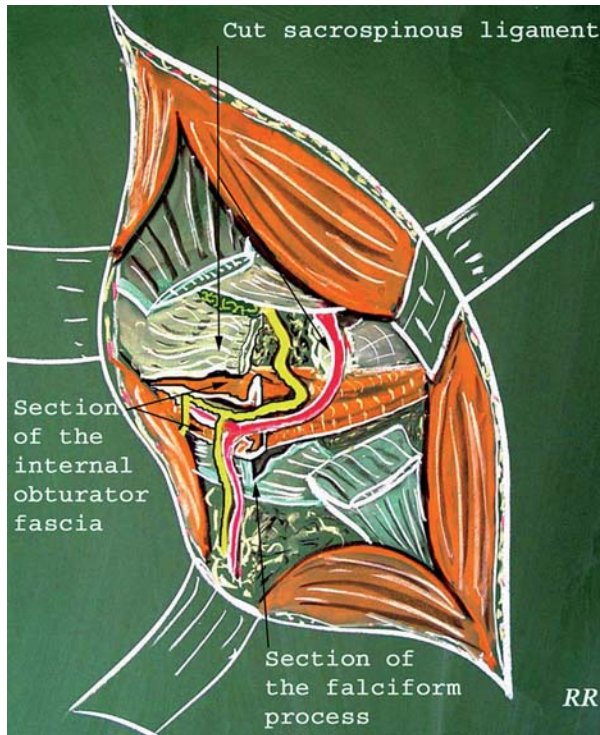
Fig. 9. The fascia of the internal obturator muscle is opened, the falciform process and the sacrospinous ligament are cut. The nerve is then transposed ventrally to the ischiatic spine. The nerve is then free all along its course

fore validated and can be reasonably proposed for perineal pain patients, according to the clinical signs mentioned above. The efficacy of the surgery confirms the etiology of these postural perineal pain syndromes previously considered as reflecting pathology within the pelvic visceras idiopathic or psychogenic.

Failure analysis: diagnostic error is always possible of course but long lasting or tight compression preoperatively could account for irreversible nerve fiber damage initiating chronic neuropathic pain. With this aim in view, the earlier people are operated on, the better the result will be. Incomplete release of the nerve by the transgluteal approach should be avoided. Recently we have positioned the patient in a genu-pectoral installation which allows a better view of the distal part of the nerve trunk.

Retrospective study

From January 1994 to December 2000, 500 patients were operated on by a single surgeon. Only 158 patients were selected, i.e. 248 nerves. The fact is that most of them came from abroad or far from Nantes.

Pain

Preop VAS was 77% which reflects very intense pain. Patients improved if the VAS decreased by 28 or more. Excellent results were considered if VAS decreased to 52 or more, no improvement for 1 mm or the same as before.

Age

Most of the patients were between 40 and 75 years old. The mean was 56. Not improved patients were 5 years older than improved ones. After 70, the success rate decreased significantly.

Sex

57% were females, 43% males. The better results in the men was not statistically significant.

Side

51% of patients had bilateral pain; 49% unilateral (51% left side, 49% right side). No difference was noted between each side. People with a unilateral pathology had a better result.

Characteristic of pain

The above mentioned clinical presentation is of course a good guide for the diagnosis. This pain is typically located in the perineal area, consisting of a burning feeling, increased with seated position. In fact, numerous patients have pain irradiation to the thigh, the abdominal wall, the pubis, complaints about sensation of a foreign body in the rectum or vagina, have pain even when standing or at night when lying. The results are the same even if the clinical presentation is atypical.

Time for evaluation

We must wait at least one year after surgery to evaluate the pain on the VAS score. Patients are informed that the definitive result will not be appreciated sooner, in spite of some early improvements.

Time for surgery since the beginning of pain

66% of patients suffering for less than 6 years improved versus 40% after 7 years duration of pain. This is an important point. Diagnosis must be made much earlier if we want to improve the results.

Neurophysiological data

The distal motor latency is, in our experience, the best test to confirm that the pudendal nerve is entrapped. Normal score is less than 5 milliseconds. When the score is more than 9 milliseconds, results are poor.

Blocks

They constitute a therapeutic weapon and a diagnostic test.

When the preoperative anaesthetic block at the level of the ischiatic spine is negative, only 17% of patients improve. If the block is positive and a per operating conflict is found, 65.5% of patients improve. If no conflict is found at this level, only 50% improve. Among them, 50% had a conflict at the level of Alcock's canal.

59% of patients with a positive diagnostic block at Alcock's canal improved. Among them, 91% had also a conflict at the level of the ischiatic spine and a positive diagnosis test at this level.

The steroid preop blocks seem to persist longer in improved patients.

Origin of pain

67% of post traumatic pain improved which is a little better than spontaneous neuralgias.

Peri-operative findings

They are very important to consider. Normal anatomy leads to a bad result as mentioned above. To summarize, the best condition is to find a flattened nerve with a conflict at the level of the ischiatic spine only. 74% of these patients improve. If the entrapment is noted in Alcock's canal, only 50% improve. If the entrapment is at the two levels, the success rate is 60%.

Results

Finally, fewer than two third of patients improve after the surgery, nearly one third stay in the same condition, 1% feel worse than before.

Complications of the surgery

We never encountered any hypoesthesia nor motor complication after surgery. There was less than 1% of hematomas and sepsis. Less than 1% of the patients felt worse after surgery. Most of them estimated their pain at 100 on the VAS prior to surgery.

Comments

The results of this retrospective study pointed out some failures for nerves entrapped in Alcock's canal. To obtain a good distal decompression we defined

a new preoperative position i.e. in genu pectoral. It allows us to enter deeply into the canal and to cut the fascia of the internal obturator muscle under visual control. With the same aim, we have started to cut the nerve to this muscle to obtain a definitive amyotrophy reducing the pressure in the tunnel. We will publish our results later.

The prospective randomized study validated the surgery. It must be clearly keep in mind that before our clinical work, the patients were considered as psychiatric. Blocks and surgery have transformed the life of patients from a terrible social condition in two third of cases.

These results could be considered rather poor, but are consistent with results obtained in the surgery of the spinal lumbar disc for example. If we consider that the blocks definitively improve about 50% of patients and that among them 65% improve after surgery, only 17% of the patients we treat failed to improve.

Other diagnoses

Symptomatic pain provoked by acute dermatological infections of the perineum are easy to diagnose and leads to symptomatic treatment.

Pain persisting at night suggests a tumour to be excluded radiologically. Other neuropathic pains must be explored and analysed in their context. Surgery requiring trans abdominal or pelvic wall approaches may cut sensory nerves such as the ilio-hypogastric, ilio-inguinal or genito-femoral trunks [8, 16, 21, 30]. They invade partly the supplying area of the pudendal nerve. The patient's story is clear enough and a trigger zone must be sought (Tinel's sign) leading to the diagnosis of a post traumatic neuroma.

The possibility of radicular pain arising from orthopaedic problems at the thoraco-lumbar area must be considered and can be treated by physiotherapy and specific blocks [18].

Testicular [9] and urethral [36] pains must be carefully analysed. Their nervous supply arises from the autonomic system which is situated at the thoraco-lumbar junction. Specific blocks may be helpful.

Vestibulodynias (vulvar pain) are very special [12]. Women suffer from an allodynia which is the main sign and any local contact is unbearable.

Bladder pain is frequently encountered during our consultations. The pain increases during the filling up of the bladder and stops after the micturition.

Proctalgia fugax is a paroxysual pain in the anal region during the night, awaking the patients, leading even to syncope and resolves within half an hour. Its origin is unknown.

Some patients may complain about a sciatic pain without back ache which can be the main pain or in association with a perineal pain. Different presentations occur: a sciatic pain may reach only the posterior aspect of the buttock and of the thigh. A compression of the posterior cutaneous nerve of the thigh

can be provoked. The entrapment is caused by the piriformis muscle [10, 33] which can damage both the posterior cutaneous muscle of the thigh and the pudendal nerve. A sciatica which reaches, without any lumbar pain, the inferior limb until the foot may be due to a compression of the anterior part of the ischiatic trunk and/or the pudendal nerve by the internal obturator muscle [19]. A deep gluteal syndrome has been described when the two muscles are involved [17]. Physiotherapy focussed on the stretching of these muscles, specific blocks, and even surgery may help.

A pain situated laterally to the anal region when sitting could be explained by a compression of the inferior clunial nerve. Patients with this pathology complain of pain when sitting on a hard seat. Pudendal pain arises when seating on a soft seat most of the time.

The obturator neuralgia is different, being located away from the perineum and reaching the superior and medial aspect of the thigh. Walking or stepping on one foot is then painful. Herniations must be evoked [20]. A real entrapment syndrome has been established [31]. We have one case of such a pain after surgery for urethral incontinence (unpublished observation).

Coccydynias start in the coccygeal area, back from the anal region, occur when sitting and when standing up and then become very acute. People therefore sit in a bending position avoiding the compression of the coccyx. A radiological instability may be demonstrated by dynamic X-rays in sitting and standing position. Clinically, intra rectal pressure brings on the pain when moving the coccyx. The instability, if found is between the coccygial vertebre. Blocks may be positive. Surgery is rarely required.

Herpetic infection especially in women may later induce chronic pain which is not influenced by any position and persists during the night.

Chronic prostatitis [14] is a fashionable syndrome but, unfortunately, the bacteriological proof is rare. Very often a true pudendal pain may be called prostatitis.

Sacral root pains are quite different. They are mostly accompanied by lumbar pain, and reflexes are absent. Anesthesia is noted and mild motor disturbances are frequent.

Conclusion

Perineal pain constitutes a difficult diagnostic problem. Nevertheless, patients may describe a typical history of burning pain in the territory of the pudendal nerve when sitting. An entrapment syndrome of the pudendal nerve must be diagnosed and complementary investigations must be done to be sure that there is no other explanation for the pain.

Neurophysiological tests are performed, searching for an increase of the distal motor latency. Blocks are proposed under CT scan control which are at

the same time a diagnostic test and a therapeutic weapon. Surgery is then performed if the pain is still present. The transgluteal approach allows the release of all compressive factors. Nearly two thirds of patients improve after surgery. Long duration of the illness, advanced age of the patients, are predictive factors, and must be reduced by an earlier diagnosis.

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