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Entrapment of the posterior femoral cutaneous nerve and its inferior cluneal branches: anatomical basis of surgery for inferior cluneal neuralgia

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Abstract
Purpose The apparent failure of pudendal nerve surgery in some patients has led us to suggest the possibility of entrapment of other adjacent nerve structures, leading to the concept of inferior cluneal neuralgia. Via its numerous collateral branches, the posterior femoral cutaneous nerve innervates a very extensive territory including the posterior surface of the thigh, the infragluteal fold, the skin over the ischial tuberosity, but also the lateral anal region, scrotum or labium majus via its perineal branch.
Methods We described the pathophysiological features of cluneal neuralgia, the surgical technique and our preliminary results.
Results We performed a transmuscular approach leading to the fat of the deep gluteal region. Exploration was continued cranially underneath the piriformis, looking for potential entrapments affecting the posterior femoral cutaneous nerve and the sciatic nerve. Nerve decompression on the lateral surface of the ischial tuberosity was then performed. A constant anatomical finding must be highlighted: the presence of a lateral fibrous expansion from the ischium passing behind the nerves and vessels, especially the posterior femoral cutaneous nerve and its perineal branches. In our patients, release of this expansion allowed decompression of the nerve trapped by this expansion.
Conclusion Cluneal neuralgia constitutes a distinct entity of perineal pain, which must be identified and distinguished from pudendal neuralgia. Surgery should be performed via a transgluteal approach. A lateral ischial obstacle must be investigated, in the form of a constant fibrous expansion, which, like a retinaculum, can cause nerve entrapment.

Keywords Surgical · Operative technique · Posterior femoral cutaneous nerve · Cluneal · Neuralgia · Transgluteal approach

Introduction

The apparent failure of pudendal nerve surgery in some patients [7] has led us to suggest the possibility of entrapment of other adjacent nerve structures, leading, on the basis of anatomical studies, to the concept of inferior cluneal neuralgia, the terminology of which will be explained herein [1, 8]. Some patients undergoing pudendal nerve surgery may experience persistent pain responsible for the same clinical and social repercussions, but clinical interview revealed that the nature of this pain had changed and these patients no longer presented the typical characteristics of classical pudendal neuralgia [6]. These patients no longer experienced anal pain, but lateral anal and medial ischial pain sometimes extending into the infragluteal fold or even onto the posterior surface of the thigh, while penile, clitoral or anal pain had resolved [8]. The contribution of positional discomfort to the patient's disability was just as important as in the previous pudendal neuralgia [10]. The most obvious conclusion was therefore failure of pudendal nerve surgery, but this was certainly not the case, as the patient's perineal pain, justifying pudendal nerve surgery, had resolved. In this paper, we describe the features of cluneal neuralgia, the surgical technique and our preliminary results.

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Anatomical data

Via its numerous collateral branches, the posterior femoral cutaneous nerve innervates a very extensive territory including the posterior surface of the thigh, the infragluteal fold, the skin over the ischial tuberosity, and also the lateral anal region, scrotum or labium majus [10]. An anatomical study demonstrated that the perineal branch of the posterior femoral cutaneous nerve is one of the many inferior cluneal nerves [1].

Anatomical descriptions underlie the surgical approach of the inferior cluneal nerves described herein. Fifty cadavers were examined bilaterally between September 2014 and June 2016. The posterior femoral cutaneous nerve classically arises from the sacral plexus, predominantly from the S2 nerve root. It then accompanies the sciatic nerve, usually coursing medially or posteriorly to this nerve. The posterior femoral cutaneous nerve gives rise, medially, to inferior cluneal nerves, which course below the inferior border of the gluteus maximus muscle and enter the skin of the buttocks, providing sensory innervation to this region. The perineal branch generally arises from the medial border of the posterior femoral cutaneous nerve, on the lateral aspect of the ischial tuberosity and then courses subcutaneously over the origins of the hamstring muscles, transversely crossing over the tendons lateromedially. The perineal branch then innervates the lateral anal, scrotal or labial skin [1].

During the dissections, care was taken to the level of the birth of the cluneal branches in relation to the inferior edge of the ischial tuberosity. The posterior femoral cutaneous nerve always appears lying against the lateral aspect of the ischial tuberosity (Fig. 1). For all cadavers, inferior cluneal nerves stem from the medial edge of the posterior femoral cutaneous nerve, a few centimetres from its origin, at a maximum of 3 cm under the inferior edge of the ischial tuberosity (Figs. 2, 3). Cluneal nerves therefore provide sensory innervation to the mid-ischial, lateral anal and scroto-labial cutaneous territory; with the exception of the scrotum or labium majus, the territory of the inferior cluneal nerve is therefore different from that of the pudendal nerve.

The presence of a lateral fibrous expansion from the ischium, which did not appear threatening in cadavers, was in fact much tauter and possibly compressed the nerves in patients. This fibrous expansion constitutes the principal site of entrapment of the nerve and we should emphasise that it was examined in all cadavers. This fibrous expansion, thicker on its ischial insertion than laterally, spread out between the ischial tuberosity and the deep gluteal fascia. It seemed to lengthen the distal insertion of the sacrotuberal ligament. Moreover, the entrapment is situated at this level on the medial part of

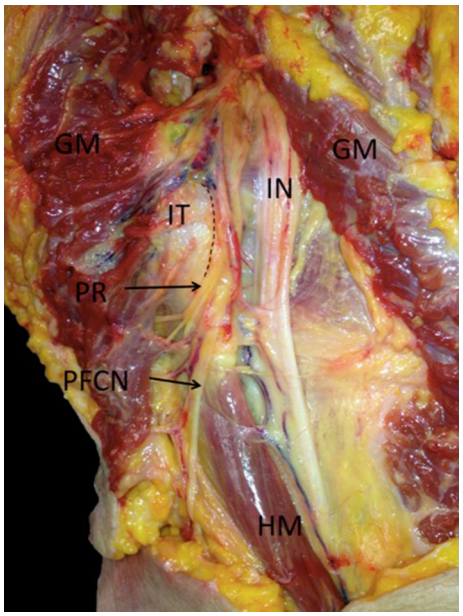

Fig. 1 Right side. The posterior femoral cutaneous nerve is located on the lateral side of the ischial tuberosity. The gluteus maximus muscle is incised (unembalmed cadaver). *GM* gluteus maximus incised, *IT* ischial tuberosity, *IN* sciatic nerve, *PFCN* posterior femoral cutaneous nerve, *HM* hamstring muscles, *PR* perineal ramus, *dashed line* lateral edge of the ischial tuberosity

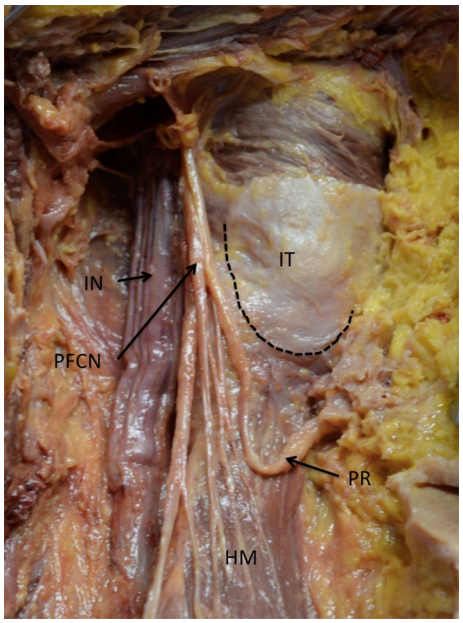

Fig. 2 Left side (embalmed cadaver). The posterior femoral cutaneous nerve is located medially and dorsally to the sciatic nerve. The perineal branches emerged from the posterior femoral cutaneous nerve at the level of the inferior extremity of the ischial tuberosity. *IT* ischial tuberosity, *IN* sciatic nerve, *PFCN* posterior femoral cutaneous nerve, *HM* hamstring muscles, *PR* perineal ramus, *dashed line* lateral edge of the ischial tuberosity

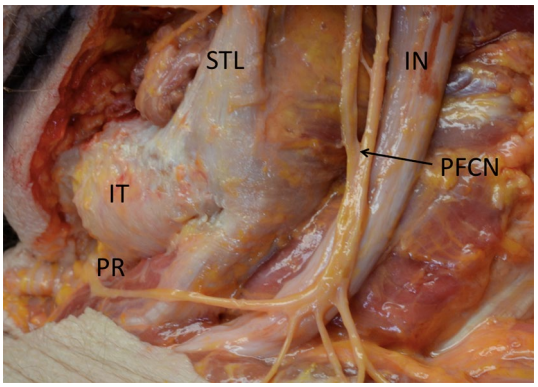

Fig. 3 Right side (unembalmed cadaver). The posterior femoral cutaneous nerve is located medially and dorsally to the sciatic nerve. The perineal branches emerged from the posterior femoral cutaneous nerve at the level of the inferior extremity of the ischial tuberosity. *IT* ischial tuberosity, *IN* sciatic nerve, *PFCN* posterior femoral cutaneous nerve, *PR* perineal ramus, *STL* sacrotuberal ligament, *dashed line* lateral edge of the ischial tuberosity

the fibrous expansion beside the lateral part of the ischial tuberosity in sitting position.

Moreover, coexisting with this entrapment, there were always adhesions between the nerve trunk and the lateral aspect of the ischial tuberosity. At this level, the lack of mobility of the nerve can, by itself, lead to pain like in other tunnel syndromes (i.e. ulnar nerve on the elbow).

Surgical technique

Principle Surgery must concern both the posterior cutaneous femoral nerve and its perineal branches.

Technique The subject is placed in the prone position with the thighs flexed onto the pelvis. An incision is made in the axis of the gluteus maximus muscle fibres laterally to the ischial tuberosity (Fig. 4). Dissection of gluteus maximus muscle fibres is only minimally traumatic, as the fibres are separated in their long axis. The passage through the muscle is complete when the fat of the deep gluteal region appears, which is constantly present around the nerves. This region corresponds to that of the trochanteric muscles. The location of the entrapment is assessed by digital exploration from the infrapiriformis tunnel to the fibrous expansion caudally. The nerves are dissected at the caudal and medial margins of the piriformis muscle and, when entrapment is observed, the piriformis muscle is at least partially sectioned in the zone adjacent to the nerves. Note that this muscle can comprise a powerful medial tendon that can only be assessed by palpation, as it is generally covered by muscle fibres. When this tendon is detected, it should be sectioned to avoid entrapment between the tendon and the nerve.

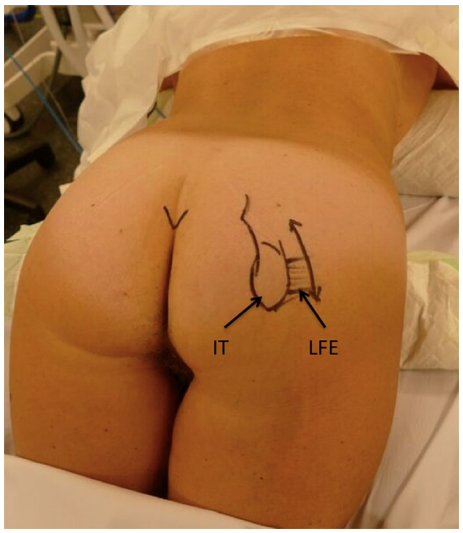

Fig. 4 Posterior view of patient positioning, showing the incision, the projection of the ischial tuberosity, which can be palpated and identified, and the lateral fibrous expansion. *IT* ischial tuberosity, *LFE* lateral fibrous expansion

Nerve decompression on the lateral surface of the ischial tuberosity is then performed and continued as far as the origin of the perineal branches, which are released by finger dissection of the origins of the hamstring muscles. Entrapments between the posterior femoral cutaneous nerve, the sacrotuberous ligament and the ischial tuberosity may be observed anatomically, but a constant anatomical finding must be highlighted: the presence of a lateral fibrous expansion from the ischium passing behind the nerves and vessels, especially the posterior femoral cutaneous nerve and its perineal branches (Fig. 5). This fibrous

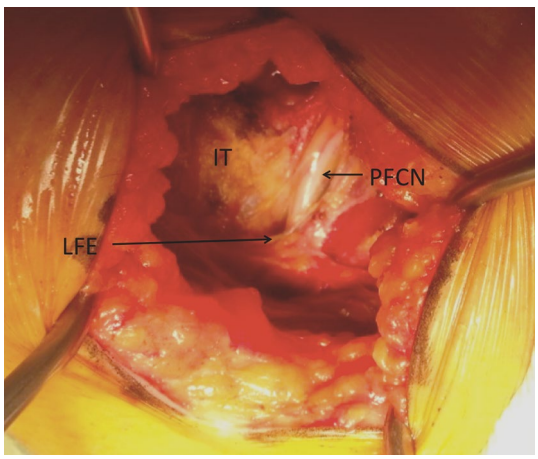

Fig. 5 Right side. Surgical illustration with schematic drawing showing the posterior femoral cutaneous nerve and the fibrous expansion. *IT* ischial tuberosity, *PFCN* posterior femoral cutaneous nerve, *LFE* lateral fibrous expansion

expansion, which has not been described anatomically to our knowledge, was constantly observed during cadaver dissections, but no entrapment was observed in cadaver specimens between this band and the nerve. In contrast, in our patients, release of this band allowed decompression of the nerve trapped by this band. This fibrous expansion was observed in all surgical procedures performed on suffering patients leading to macroscopic compression of the nerve.

In patients with combined pudendal and cluneal neuralgia, a single incision is made on either side of a transverse line through the summit of the coccyx over the sacrotuberous ligament. Surgery is initiated via a pudendal approach in the superomedial part of the incision, at the site of "hourglass" narrowing of the sacrotuberous ligament [5]. Pudendal nerve decompression is performed according to a previously published technique [7]. Dissection is then simply extended caudally to the sacrotuberous ligament, which is resected as far as the ischium. The posterior femoral cutaneous nerve and the inferior cluneal branches are then exposed via a lateral approach, as described above.

Discussion

Cluneal neuralgia constitutes a distinct entity of perineal pain, which must be identified and distinguished from pudendal neuralgia. Clinical interview is the main element of the diagnosis and description of the painful zone constitutes the cornerstone of this diagnosis. The features of pudendal neuralgia will not be described in detail here, as they are well known, but several points should be emphasised. This pain can involve the perineum, but exclusively the scrotum or labium majus, a territory shared with the pudendal nerve and the ilioinguinal nerve as well as other nerves, as we have demonstrated anatomically [1]. In particular, cluneal neuralgia is more lateral than pudendal neuralgia and does not involve either the anus or the penis or the clitoris. It can be accompanied by pain in the territory of the posterior femoral cutaneous nerve and/or its lateral inferior cluneal branches, which explains the possible distribution of this pain in the infragluteal fold, genitofemoral fold, ischial region, and posterior surface of the thigh. This pain is generally not accompanied by any autonomic disorders. A transient response to local anaesthetic nerve block is necessary to propose surgery [3, 4]. There is no limitation of the technique if the clinical interview is indicative and the anaesthetic nerve block positive.

The name of the inferior cluneal nerve is actually an over-simplification, as these branches of the posterior femoral cutaneous nerve actually correspond to a number of nerves. This over-simplistic terminology allows pudendal neuralgia to be distinguished from inferior cluneal neuralgia. Note that several perineal branches may be present.

Only the transgluteal approach, which has been shown to be a valuable approach for pudendal nerve surgery [9], can allow surgical management of these various problems, especially in patients with multiple nerve entrapments. Other surgical approaches were described, notably for the treatment of neuroma of the posterior femoral cutaneous nerve [2], but we think that our proposed surgical technique has two main advantages: it can confirm the definitive diagnosis by demonstrating anatomical entrapment of the various nerves and it allows decompression of all nerves involved via a single incision. Moreover, this approach is devoid of any complications, apart from postoperative haematoma or abscess, which are nonspecific and extremely rare in this setting.

A total of 85 patients were operated for cluneal neuralgia over a 1-year period between September 2013 and 2014 in our centre with the technique described above. This technique is now commonly used in our centre. All patients were reviewed 6 months after surgery. Evaluation of the improvement of painful symptoms was expressed as the percentage improvement at 6 months. Among these 85 patients, 24 were operated on for isolated cluneal neurolysis (16 unilateral nerves and 8 bilateral nerves) and 61 were operated on for both pudendal and cluneal nerves (43 concerned combination of unilateral pudendal and cluneal neuralgia and 18 concerned combination of bilateral pudendal and cluneal neuralgia). According to our experience, among the patients operated for perineal pain, cluneal nerves were involved in about 25% of cases and women seem to be more concerned.

Based on current estimates, the results of surgery for cluneal neuralgia are identical to those obtained with pudendal nerve surgery: in total, 70% of patients were improved or completely cured, while 30% were persistent surgical failures [7]. As in the case of pudendal neuralgia, it often takes 5–6 months to observe the first signs of improvement.

Conclusion

Anatomical studies demonstrate the topography, distribution, cutaneous territories and anatomical relations of the posterior femoral cutaneous nerve and its cluneal branches involved in a well-defined nosological framework. When the diagnosis has been established, surgery may be indicated after the failure of medical treatment. This surgery is essentially anatomical. Surgery should be performed via a transgluteal approach following a specific diagnostic nerve block in addition to the clinical criteria for surgery defined for pudendal neuralgia. A lateral ischial obstacle must be investigated, in the form of a constant fibrous expansion, which, like a retinaculum, can cause nerve entrapment. In patients with a combination of cluneal neuralgia and

pudendal neuralgia, the one-step transgluteal approach via the same incision is performed, allowing simple access to the various sites of entrapment. The preliminary results obtained with this technique are promising.

Compliance with ethical standards

Conflict of interest The authors declare no conflict of interests in relation to this article.

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