Intraneural ganglia in the hip and pelvic region

Clinical article

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Object. The authors describe their experience in a series of cases of intraneural ganglia within the hip and pelvic regions, and explain the mechanism of formation and propagation of this pathological entity.

Methods. Five patients with 6 intraneural ganglia are presented. Four patients presented with symptomatic intraneural ganglia in the buttock and pelvis affecting the sciatic and lumbosacral plexus elements. An asymptomatic cyst affecting the opposite sciatic nerve was found on MR imaging in 1 patient. The fifth patient, previously reported on by another group, had an obturator intraneural ganglion that the authors reinterpreted.

Results. All 5 intraneural ganglia affecting the sciatic and lumbosacral plexus elements were found to have a joint connection to the posteromedial aspect of the hip joint; the obturator intraneural cyst had a joint connection to the anteromedial aspect of the hip joint. In all cases, initial review of the MR images led to their misinterpretation.

Conclusions. To the authors’ knowledge, these are the first cases of intraneural ganglia demonstrated to have a connection to the hip joint. This finding at a rare site provides further evidence for the unifying articular (synovial) theory for the formation of intraneural ganglia and reveals a shared mechanism for their propagation. Furthermore, understanding the pathogenesis of these lesions provides insight into their successful treatment and their recurrence. (DOI: 10.3171/2009.2.JNS081720)

Key Words • sciatic nerve • articular (synovial) theory • cyst

Intraneural ganglia are relatively rare cysts located within the epineurium of peripheral nerves. Despite a long and controversial history regarding their formation, recent evidence has substantiated an articular (synovial) origin, particularly for the prototype peroneal nerve at the fibular neck region. To explain the formation and propagation of these intraneural cysts, a unified theory has been proposed. The principles of this theory include: 1) an articular branch connection to a synovial joint through a labral/capsular rent; 2) intraneural propagation along a path of least resistance; and 3) dynamic pressure fluxes influencing the size and direction of cyst propagation. This unified theory also explains the more frequent extraneural ganglia that occur when capsular rents are apart from articular branches, leading to nonneural pedicles; these extraneurally extending cysts may occasionally compress (or adhere) to a nerve, causing neuropathy. Because they are not constrained by epineurium, these extraneural cysts tend to be more globular than the more tubular intraneural cysts. The 2 types of cysts may coexist or one may arise after treatment for the other, reflecting the shared joint pathology.

The finding of a joint connection was popularized by the prototype peroneal nerve intraneural ganglion, which arises at the anterior portion of the superior tibiofibular joint, but substantiated at other rare sites. A consistent articular connection has been corroborated by other authors. When intraneural cysts occur at unusual sites, their joint connection may be difficult to establish, especially if they occur far from synovial joints. Identification of this joint connection is important in that failure to recognize and treat it frequently leads to intraneural recurrences.

To our knowledge, an intraneural cyst arising from the hip joint has not been previously reported. We hypothesized that new cases of intraneural ganglia affecting the sciatic nerve and lumbosacral plexus as well as a previously reported case of an obturator intraneural
cyst would arise from the hip joint. The description of these former cysts validates the term “sciatic intraneural ganglia.” Because of the variable origin of the articular branches, these lesions may be either sciatic or lumbosacral plexus cysts. Previously, the entity of a “sciatic intraneural ganglia” was considered a misnomer when it described a peroneal or tibial intraneural ganglion cyst derived from the superior tibiofibular joint and extending proximally to the sciatic nerve; rare examples of this could extend to the level of the buttock and should not be confused with cases in this report originating from the hip and extending to the pelvis.

The purpose of this report was to clarify a few remaining controversies and generalize on previous observations made at other joints, including: 1) that such intraneural cysts can arise in the vicinity of the hip; 2) that these intraneural cysts arise from the hip joint (consistent with the articular theory), even when identified far from that joint; 3) that common propagation patterns can be discerned; 4) that analogies to similar joints, notably the shoulder joint (upper extremity counterpart of the hip joint) can be drawn; and 5) that all nerves giving rise to articular branches to the hip joint could potentially be affected by an intraneural cyst. Support for the articular theory for these intraneural cysts (sciatic, lumbosacral plexus, and obturator) at an unusual site (hip) has important implications for treatment of this lesion and the prevention of recurrences.

Methods

All cases of intraneural ganglia in the hip and pelvic region known to the primary author (R.J.S.) were included in this study. Each case was reviewed for the available clinical information, initial MR imaging interpretation, operative treatment, and postoperative results. Each MR imaging study was then retrospectively reinterpreted by a single musculoskeletal radiologist (K.K.A.) experienced in the imaging features of intraneural ganglia.

Clinical Information

Materials from 5 patients with 6 intraneural ganglia were reviewed: 4 patients (Cases 1–4) had 5 sciatic and lumbosacral plexus intraneural cysts, and 1 patient (Case 5) had an obturator intraneural cyst. Three men and 1 woman (26, 33, 53, and 55 years of age) with sciatic intraneural ganglia presented with symptoms and signs of sciat ic neuropathy that had occurred over months to years (range 2 months to 2 years). On examination, 2 patients (Cases 2 and 4) had peroneal (predominant) and tibial dysfunction, 1 had peroneal dysfunction (Case 1), and 1 had a normal examination (Case 3). The Tinel sign was present at the buttock level in the patient in Case 4, and a mass lesion could not be detected on examination in any of the patients. Electromyography was performed in 2 cases: the results were normal in the patient with a normal physical examination (Case 3), peroneal neuropathy at the knee was found in 1 patient (Case 2) in whom a lumbosacral plexopathy subsequently developed.

The patient in Case 5 had an obturator intraneural ganglion that had been previously reported on by another group. This 71-year-old man presented with right leg pain that had persisted for several months. No neurological examination was reported, and no electromyography was done.

Imaging Review

In the 4 patients (Cases 1–4) with sciatic and lumbosacral plexus cysts, initial interpretation of MR images demonstrated a cystic lesion within the sciatic notch or pelvis. In no case was the joint connection identified prospectively on the initial interpretation. In 1 of these patients (Case 1), a joint connection was identified on MR arthrography. The obturator intraneural cyst (Case 5) was noted to extend into the obturator foramen on the initial MR imaging interpretation, but a joint connection was not appreciated.

In our retrospective review of the imaging studies in these patients, we found that, when reviewed collectively, all of them had remarkably similar findings: 1) evidence of a joint connection; 2) abnormalities at the level of the hip joint including degenerative joint disease; and 3) preferential (proximal) ascent of the intraneural cyst (Figs. 1–5).

Sciatic Nerve/Lumbosacral Plexus Cases (Cases 1–4)

Cyst ascent within the articular branch could be demonstrated in each case originating from the posteromedial aspect of the hip joint. The cyst within the articular branch itself was smaller, no more than 5 mm in maximum width, compared with the balloon-like expansion of the cyst at the level of the sciatic notch (where crossover at the level of the sciatic nerve and/or lumbosacral plexus occurs). The cysts extended proximally in each case to the level of the sciatic notch. In 1 patient (Case 4), there was questionable extension into the pelvis; in the other 3 patients (Cases 1–3), the cyst extended well into the pelvis and nearly to the level of the origin of the lumbosacral plexus, preferentially involving L-4 and L-5 to the extent that this could be discerned on these studies (Fig. 3B). Distal to the point of crossover, there was subtle evidence of cyst in all cases within the sciatic nerve, indicative of descent (Fig. 2). There was, however, no direct extension from the hip joint to the sciatic nerve at the level of the buttock and hip joint. In the patient (Case 1) in whom high-resolution MR arthrography images were available, cyst origin from a labral tear was clearly seen (Fig. 2): contrast material was observed extending from the tear into the cyst with clear evidence of contrast within the articular branch from the hip joint. Contrast was not seen in the more proximal cyst involving the sciatic nerve. We believe, based on our prior experience, that contrast could have been seen within this portion of the cyst with provocative maneuvers (exercise) that would have raised intraarticular pressure.

In our retrospective MR imaging review, the patient in Case 2 was found to have bilateral intraneural cysts involving the sciatic nerve, which had not been identified on initial image interpretation (Fig. 3). Only the side on which the cyst extended into the pelvis was symptomatic.
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in this patient, whereas the opposite side extended to the sciatic notch.

Subtle denervation changes involving the quadratus femoris and inferior gemellus muscles were present in all cases. In the patient (Case 1) where this was best appreciated, the changes in the quadratus femoris muscle included some evidence of fatty atrophy and were more prominent than in the inferior gemellus. A single case (Case 2) had patchy signal changes and atrophy within a portion of the gluteus medius muscle on the affected side.

Obturator Nerve Involvement (Case 5)

Our review of the original MR images suggested that the lesion in the patient in Case 5 was intraneural, and confirmed the joint-related nature of the cyst from the anteromedial aspect of the hip (Fig. 5). This cyst also exhibited proximal ascent.

Operative Procedure

Of the 4 patients with sciatic and lumbosacral cysts, 2 (Cases 1 and 4) underwent a posterior transgluteal exploration and cyst evacuation and 1 (Case 2) an anterior transabdominal approach. In the patient (Case 1) in whom a joint connection was appreciated on preoperative MR arthrography, the cyst was decompressed at the buttock level, but the joint connection was not appreciated intraoperatorically. The cysts in the patients in Cases 1, 2, and 4 were thought to have been evacuated and partially resected. In a fourth patient (Case 3, Fig. 4), the hip lesion was treated with an offset procedure and labral repair, and the intraneural cyst was not decompressed.

A pelvic approach was used in the patient with the obturator intraneural cyst (Case 5). In this case, the nerve and the cyst were resected along with the obturator artery and vein, and its joint connection was neither identified nor treated.

Postoperative Course

The follow-up period was 2–36 months (mean 16 months). All patients experienced symptomatic (pain) relief to varying degrees. Partial relief was obtained in the

Fig. 1. Preoperative images obtained in a patient with a sciatic intraneural cyst. Case 1. A: Axial T2-weighted fast spin echo (FSE) image with fat suppression from an MR arthrogram shows fluid signal within a posterior paralabral cyst (arrow) at the hip with cyst extension medially into the articular branch. B: Axial T1-weighted FSE image with intraarticular contrast at the same level shows contrast-enhancement within the paralabral cyst (arrow) and proximal articular branch. C: Axial T2-weighted FSE image with fat suppression slightly superior to the level of panels A and B shows fluid extending proximally within the articular branch of the nerve (arrow). Note the partial volume effect of the cyst within the sciatic nerve just medial to the acetabulum (arrowhead). D: Axial T1-weighted FSE image with intraarticular contrast shows contrast within the more proximal articular branch of the nerve (arrow). E: Coronal proton density-weighted FSE image shows balloon-like cyst expansion within the lumbosacral plexus (asterisk) with streak-like continued ascent within the lumbosacral plexus (arrow). F: Coronal maximum intensity projection from a T2-weighted FSE data set shows the full extent of the cyst from its joint origin extending proximally from the hip joint to the intrapelvic sciatic nerve and lumbosacral plexus (dashed arrow). Note expansion of the cyst within the lumbosacral plexus (asterisk) and distal extension into the sciatic nerve (arrowhead).
patients in Cases 2–4, and complete relief in Cases 1 and 5. There was partial clinical motor function improvement in all cases involving the sciatic nerve; the patient with the intraneural obturator nerve cyst experienced postoperative impairment of adduction and partial sensory loss in the inner thigh distribution, as expected from the resection of the obturator nerve.31 Only 2 patients (Cases 1 and 3) underwent postoperative MR imaging. In the patient with a sciatic intraneural cyst in whom the joint disorder was addressed primarily (Case 3), there was no evidence of recurrence on MR images obtained 2 years postoperatively (Fig. 6). In the other patient (Case 1) in whom the joint connection was identified on MR arthrography but not intraoperatively, MR imaging done 2 months postoperatively revealed evidence of an intraneural cyst within the articular branch that extended to the level of the sciatic notch (Fig. 7). The patient nevertheless showed clinical improvement despite intraneural cyst persistence and/or recurrence.

**Discussion**

Our results confirmed that intraneural ganglia affecting the sciatic nerve, lumbosacral plexus (sciatic complex), and obturator nerve have a joint origin, the hip, consistent with the unified articular (synovial) theory. When collectively presented, the similarities in these cases attest to the relative consistency of the anatomy and the shared mechanism of formation and propagation. Despite a common disease process explaining the similar imaging features, the true nature of this pathological entity was not appreciated prospectively in any of the individual cases.
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Fig. 4. Preoperative images obtained in a patient with a sciatic intraneural cyst. Case 3. A: Axial T2-weighted FSE image of the pelvis with fat suppression shows a cyst (arrow) arising from a tear of the posterior labrum of the hip. Note posterior extension of the cyst along the acetabulum indicating intraneural ascent. B: Axial T2-weighted FSE image with fat suppression slightly superior to A demonstrating more medial intraneural cyst extension along the posterior acetabulum within the articular branch (arrows). C: Oblique coronal T2-weighted FSE image with fat suppression at the level of the sciatic notch showing intrapelvic extension of the intraneural cyst (asterisk). Note the adjacent sciatic nerve which is slightly hyperintense (arrow). D: Sagittal maximum intensity projection from a sagittal T2-weighted FSE data set showing the cyst arising from the posterior labral tear (arrow) with posterior-superior extension within the articular branch of the nerve (asterisks).

Fig. 5. Preoperative images obtained in a patient with an obturator intraneural cyst (patient previously reported in the study by Uchida et al., 2006). Case 5. A: Coronal T2-weighted FSE image demonstrates the joint connection (arrow) to the anterior hip with intraneural cyst (asterisk) extending along the obturator internus muscle within the pelvis along the course of the articular branch of the obturator nerve. B and C: Axial T2-weighted FSE images demonstrate the joint connection from the anterior hip joint (B, arrow) to the cyst extending along the articular branch (C, arrow) to the obturator nerve (B and C, asterisk). Joint fluid is present secondary to degenerative joint disease. Panel A is modified with permission from Wiley. The original figure appeared as Fig. 1 in Uchida A, Horiguchi A, Ide H, Hatakeyama N, Yoshimura I, Ogawa Y: Mucoid pseudocyst of the obturator nerve. Int J Urol 13:2006, p 471.
Collective retrospective review of the imaging of multiple cases of intraneural cysts at this rare site importantly permitted the visualization of a joint connection to the hip in all cases. One case of sciatic intraneural cyst was previously identified in the literature. The report of Juglard and colleagues of a hip-related cyst was indeterminate as to the specific cyst type (that is, intraneural or extraneural). In addition, their report contains early generation MR images with a single axial image of a large cyst at the sciatic notch that resembles those illustrated in our cases. In both cases a joint connection—which we believe existed—was not recognized. Our search also revealed 2 historical reports of a hip-related intraneural cyst within the femoral (crural) nerve: one was probably a cystic nerve sheath tumor in the setting of neurofibromatosis, and it is debatable whether the other was an intra- or extraneural ganglion cyst. No joint connection was detected.

The description of a major joint being affected by intraneural cysts represents an opportunity to study this entity further, and confirms that the propagation pattern of hip-related intraneural cysts follows the same principles as for the well-described cysts affecting the peroneal nerve. Although joint connections could be established on MR imaging, arthrography was used to confirm the presence of a cyst–joint communication and labral tear in the patient in Case 1. It may be important to fully evaluate the hip joint for pathology (arthritis, labral tears, impingement problems) as the primary pathology is intraarticular. The degenerative joint changes apparent in our patients and in literature reports reflect the un-
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The predominant cyst propagation was in the proximal direction (ascent) with relatively limited descent. The crossover phenomenon\(^2\) (in which the cyst in 1 neural pathway may fill a shared epineurial sheath at a junction of 2 nerves to fill another) explains the finding of multiple interconnected cysts herein in the lumbosacral plexus (Fig. 8). Clearly, experience with additional cases of such rare cysts and higher resolution scanning are necessary to refine the ability to define the specific articular branch affected and identify the nuances of crossover.

The cases presented in this study strengthen the argument that intraneural cysts and their more common extraneural counterparts are part of the spectrum of paralabral cysts of the hip joint; depending on the sites of the capsular rents to the articular branch, either one or both can be created. Extraneural ganglia are becoming increasingly associated with labral cysts,\(^9\) which in turn have been found to be associated with bone abnormalities.\(^{33}\) When these extraneural cysts occur near nerves, they have been described as producing sciatic, femoral, and obturator nerve compression.\(^{27,19,20,28,35}\) In the patient who underwent MR arthrography, the cyst was visible arising from a labral tear. The tear itself was extensive, extending both anterior and posterior, and reminiscent of a superior labrum from anterior to posterior (SLAP) tear at the shoulder with a paralabral cyst. In the cases...
reported herein where intraarticular contrast enhancement was not given, the common underlying pathological entity is also probably a labral tear with intraneural extension from a paralabral cyst. In both intraneural and extraneural cases the tear may not be perceived on imaging studies because of the lack of intraarticular contrast, which increases sensitivity compared to conventional imaging for labral lesions.28

In some cases, these masses have been misidentified as nerve sheath tumors on imaging.2 The judicious use of contrast enhancement along with close attention to MR imaging features should readily allow distinction between these 2 very different lesions. Benign nerve sheath tumors have a characteristic imaging pattern and oval shape oriented along the nerve; they also readily show enhancement after contrast injection, confirming their solid nature. Plexiform lesions will similarly enhance as solid masses and will show a fusiform, not tubular, distribution along the involved nerve. Cystic lesions will only show fine peripheral contrast enhancement and are generally oriented along the nerve in a tubular fashion. Misidentification of hip-related intraneural cysts as nerve sheath tumors on imaging studies is most commonly the result of a lack of intravenous contrast dye administration.

The retrospective evaluation of imaging in our cases has inherent limitations: 1) optimized technical parameters may not have been used for image acquisition; 2) the imaging study may be incomplete in terms of contrast administration; and 3) the planes available for review may be insufficient. In most of the cases reviewed, images were obtained at lower than ideal spatial resolution and with incomplete proximal coverage, which could have confirmed cyst extension to the lumbar and/or sacral nerve roots more definitively. However, with the use of an advanced postprocessing workstation and knowledge of the relevant anatomy, pertinent findings and cyst propagation patterns could be determined. Ideally, all cases should be evaluated with high-resolution, high-field (3T) MR arthrography with dedicated receiver coils that would include the area from L-3 through the lesser trochanter to assess the full extent of cyst involvement.

Because of the small number of cases and the nature of this study, specific recommendations for treatment cannot be established. However, previous techniques used to treat intraneural ganglia can be extrapolated.22 We believe that the articular branch connection should be addressed, ideally together with treatment of the hip lesion. A limited decompression of the cyst through the same buttock approach affords an immediate neural decompression. Resection of the cyst and its wall is not necessary, and resection of the nerve should be avoided. Failure to treat the articular branch can result in intraneural recurrences.

Conclusions

Hip-related intraneural ganglia, although rare, do exist. Reinterpretations of these sciatic cases and a previous obturator case support the anatomical basis of the unifying articular (synovial) theory. The imaging findings in intraneural cysts in this location are analogous to those occurring at other sites, demonstrating the consistency and reproducibility of the pathological anatomy. Understanding the mechanism of intraneural ganglia formation (particularly their joint-related origin) and propagation patterns can help formulate a surgical strategy that will improve neurological outcomes, minimize risks, and decrease the risk of recurrence. The unifying theory we described simplifies the pathogenesis and treatment of intraneural and extraneural cysts in the buttock and pelvic regions. Future experience with additional cases and high resolution imaging will undoubtedly further refine our knowledge about this entity.

Disclaimer

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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