Piriformis Syndrome (Sciatic Nerve Entrapment) Associated With Type C Sciatic Nerve Variation: A Report of Two Cases and Literature Review

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ABSTRACT

Although anatomical variation of the sciatic nerve and piriformis muscle at the greater sciatic notch is considered an important cause of piriformis syndrome, there are few reports on the surgical treatment of piriformis syndrome owing to specific anatomical variations of the sciatic nerve and piriformis muscle. In this report, we describe 2 cases of piriformis syndrome caused by a rare type C sciatic nerve variation that were surgically treated using the transgluteal approach. The first patient reported unremitting left hip and leg pain that occurred following blunt trauma to the hip. The second patient complained of chronic pain in the buttocks and right leg, which persisted even after the patient underwent lumbar fusion surgery. Severe sitting pain and sciatica are symptomatic indications for the diagnosis of piriformis syndrome. A rare “C” type sciatic nerve variation was observed on the affected side under magnetic resonance imaging. Transgluteal sciatic nerve decompression provided significant pain relief. If severe sciatic nerve deformation due to a rare sciatic nerve variation can be confirmed with typical findings of piriformis syndrome, the possibility that sciatic nerve entrapment may have occurred in this variation should be considered.

Keywords: Entrapment neuropathy; Piriformis muscle syndrome; Sciatic nerve

INTRODUCTION

Entrapment of the sciatic nerve in the deep gluteal region has long been known by the elusive name of “piriformis syndrome.” Although it was first discovered in the year 1928, the term piriformis syndrome was coined in the year 1947. However, the pathophysiology and mechanism of this condition is still not defined precisely till date. In general, it is considered as a non-discogenic cause of sciatica, which occurs due to the impingement of the sciatic nerve either through or around the piriformis muscle. It is still regarded as a controversial syndrome but is potentially common today, however, it continues to be an underdiagnosed cause of buttock pain and non-discogenic cause of sciatica.

Classically, piriformis syndrome develops due to following reasons; muscle spasm, blunt trauma to the buttck, and hypertrophy and inflammation of the piriformis muscle. However, the anatomical variations in the course of the sciatic nerve in relation to the
piriformis muscle was also considered important, because the reports of piriformis syndrome due to rare variations of the sciatic nerve associated with the piriformis muscle continued sporadically in the 1990s and 2000s. Indeed, it is a well-known fact that the sciatic nerve has various anatomical variations in the greater sciatic foramen (FIGURE 1). At the same time, there has been an increasing awareness of piriformis syndrome. In fact, several anatomical studies and magnetic resonance imaging (MRI) studies have been conducted to understand how anatomical variations in sciatic nerve lead to the development of piriformis syndrome. The prevalence of sciatic nerve variation due to piriformis muscle in the general population is known to be about 16% in cadaveric studies and literature reviews. Although it was thought to be related to the occurrence of piriformis syndrome, a recent large-scale, correlative radiographic study using 1,039 consecutive adult hip MRIs found no association between the variation of the sciatic nerve and piriformis syndrome.

FIGURE 1. Different types of anatomical relationships between the piriformis muscle and the sciatic nerve classified by Beaton and Anson. (A) Undivided sciatic nerve anterior to the piriformis muscle. (B) One division of the sciatic nerve passes through and the other division of sciatic nerve lies anterior to the bipartite piriformis muscle. (C) One division lies posterior to the piriformis muscle and the other division lies anterior to the piriformis muscle. (D) Undivided sciatic nerve passes through the piriformis muscle. (E) Multiple sciatic nerve divisions pass between the bipartite piriformis muscle. (F) One division lies posterior to the piriformis and the other division lies between the bipartite piriformis muscle. (G) Undivided sciatic nerve lies posterior to the piriformis muscle.
However, if the sciatic nerve is severely deformed due to rare variations in patients with suspected symptoms of piriformis syndrome, the probability of piriformis syndrome is considered to be high. In this study, we reported 2 cases of piriformis syndrome, which were caused by type C variation of the sciatic nerve. In these cases, the peroneal (fibula) division of the sciatic nerve passes above the piriformis muscle. On the other hand, the tibial division of the sciatic nerve passes under the piriformis muscle (FIGURE 1). Among 7 variations of the sciatic nerve, the incidence of type C variant occurs with a frequency of 0.3%–7.6%. Previous reports have described how piriformis syndrome is caused by type D and type F variations of the sciatic nerve; however, ours is the first study to report the clinical manifestations and surgical findings of piriformis syndrome, which is caused by type C variant.

**CASE REPORT**

**Case 1**

In a 38-year-old female patient, squeezing pain radiated from the left hip to the posterior thigh. The pain was chronic in nature, and it first occurred 9 months before being admitted to the hospital. At that point of time, the patient had fallen from a ladder about 1 m above the ground level. The patient’s buttocks were hit badly during the fall. As the pain in the buttock was severe, it was initially diagnosed as coccyx fracture. The patient was hospitalized for 2 weeks, but her condition did not improve. Squeezing pain continued to persist in the left buttock and the posterior thigh throughout the day. Moreover, it worsened when the patient was sitting on the chair or bed. Thus, the patient could not sit for more than 10 minutes. Several orthopedic surgeons, neurosurgeons, and pain specialists treated the patient with maximal medications, including narcotics and anticonvulsant, and epidural and caudal blocks; however, the intensity of the pain did not decrease. The patient had already undergone above/knee (A/K) amputation of the left leg when trauma was inflicted to the left lower extremity 20 years ago. At the amputation site, stump pain occurred about once a month, but the pain in the stump was occurring every day after the fall.

A physical examination of the patient was conducted in the hospital. The physicians did not come across any abnormal findings, such as sensory impairment in the lower extremities. Only mild tenderness was observed in the left sciatic notch. The piriformis stretch test could not be performed due to A/K amputation. No abnormal findings were reported in the MRI scans of the lumbar spine. No fractures were detected in the sacrum and coccyx regions when computed tomography of the pelvic bone was performed. The patient suffered from severe pain in the buttock and legs. A typical sitting intolerance was reported after the incidence of hip trauma. The patient’s severe buttock and leg pain and typical sitting intolerance following hip trauma suggested possibility of sciatic nerve entrapment of piriformis syndrome.

By performing an MRI of the hip, we investigated the condition of the sciatic nerve and piriformis muscle. The MRI scan revealed type C variation according to Beaton and Anson’s classification in the left sciatic nerve (FIGURE 2A). An exercise for the relaxation of the piriformis muscle was prescribed. The blocks of the piriformis muscle with a local anesthetic and steroids were performed. They showed a weak analgesic effect for about 1–2 hours. Medical treatment and repeated blocks were provided to treat piriformis syndrome for 2 months; however, the pain did not subside significantly. Therefore, the patient was unable to perform daily activities (6–7 out of 10 on a numerical rating scale-11 [NRS-11]). To tackle the squeezing pain, the patient was asked to undergo sciatic nerve decompression.
Sciatic nerve decompression using a transgluteal approach was performed under general anesthesia. The gluteus maximus muscle was split through an 8-cm lazy S-shaped skin incision to expose the lateral margin of the sacrotuberous ligament and the piriformis muscle. Intraoperative nerve monitoring and nerve stimulation were performed to identify the course of sciatic nerve and its peroneal and tibial divisions and to prevent nerve damage.
After identifying the inferior gluteal nerve, and the sciatic nerve and its divisions, we carefully separated the sciatic nerve from the entrapping piriformis muscle (FIGURE 2B). A surgical resection was performed on the proximal portion of the piriformis muscle, which was located below the sacrotuberous ligament. Thereafter, the peroneal division, which was located laterally, was dissected and decompressed. Then, the underlying piriformis muscle belly and the tendon were carefully separated from the underlying tibial division. Finally, we performed an external neurolysis of the sciatic nerve in these structures. A circumferential decompression of the sciatic nerve was performed on its proximal course, which extended from the sacrotuberous ligament to the lesser trochanteric level (FIGURE 2C).

Immediately after the operation, there was improvement in the tearing pain of the left buttock and leg. One month after the operation, the patient reported that the existing pain had improved by more than 70%. Moreover, the severe stump pain also disappeared after the operation. Six months after the surgery, the sitting pain no longer existed. Therefore, the drug for pain relief was discontinued because it did not interfere the daily life of the patient.

Case 2
A 62-year-old female patient presented with chronic pain in the lower back, right buttock, and right leg that did not improve even after lumbar spinal fusion surgery. The patient had a history of chronic low back pain for 10 years. Two years before back surgery, the patient suddenly developed an additional pain in her right buttock and right leg. The pain persisted throughout the day and even after the patient rested. The nature of leg pain was cold, tingling, and cramping in nature. Moreover, it worsened when numbness occurred in the lateral sole of the patient’s foot. This adverse event usually occurred while standing or walking. The pain was especially severe in the sitting position. For the past 4 years, the patient just could not sit for more than 30 minutes. Moreover, the pain persisted even when the patient laid down to rest at night. The patient was diagnosed with L3/4 retrolisthesis and lumbar stenosis. Therefore, surgeons performed posterior lumbar interbody fusion at L3/4. After undergoing lumbar fusion surgery, the patient’s lower back pain improved slightly; however, the pain did not improve at all in the right buttock and the right leg. The patient was prescribed anticonvulsants, narcotics, and lumbar epidural and caudal blocks for 2 years, but the pain was not alleviated and daily activities could not be performed by the patient.

On physical examination, there were no obvious sensory abnormality or motor weakness and the deep tendon reflex was normoactive. Tenderness was not evident in the right sciatic notch. The sitting pain of the patient was so severe that she could not sit and had to stand during the interview. Although the pace sign of the piriformis muscle was negative, the pain in the right buttock worsened while performing the flexion-adduction-internal rotation (FADIR) test. Postoperative instability, residual stenosis, or an adjacent segment degeneration was not observed in the lumbar myelographic computed tomography scan and the MRI scan of the lumbar spine. The MRI scan of the hip confirmed the development of a type C variation of the sciatic nerve. In this condition, the peroneal division of the sciatic nerve passes over the piriformis muscle (FIGURE 3A). Persistent pain radiated in the hip and leg of the patient; she had sitting intolerance and nocturnal pain. The results of FADIR test were positive. In particular, the patient showed a rare form of sciatic nerve variation. These events led to the entrapment of sciatic nerve, leading to the development of piriformis syndrome. On providing an anesthetic block to the piriformis muscle, a slight improvement was observed in the existing pain for about 2 hours. On providing 3 consecutive piriformis blocks, the patient got only temporary relief. There was no long-term improvement in the condition of the patient.
Considering the chronicity and medical intractability of piriformis syndrome caused by type C sciatic variation, decompression of the sciatic nerve through a transgluteal approach was planned. As described in case 1, a complete circumferential decompression was performed by partially resecting the piriformis muscle and the tendon encircling the sciatic nerve and its division (FIGURES 3B & C). After the operation, the numbness in the peroneal nerve persisted.
nerve existed in the right leg for 6 months. However, there was significant improvement in the severe pain of the right hip and right leg (3–4 out of 10 on NRS-11). The patient reported that the existing pain had decreased by more than half after undergoing sciatic nerve decompression. She could sit for more than one hour and had no problems while walking for one hour. Although pain in the buttock pain improved but lower back pain persisted. One year after the surgery, a spinal cord stimulator was implanted to alleviate chronic lower back pain. Thus, a significant relief was observed in the lower back pain of the patient.

**DISCUSSION**

**Piriformis syndrome**

Piriformis syndrome was originally described by Robinson in the year 1947.[17] In this syndrome, a patient presents a collection of symptoms, including severe pain in the lower back, buttock, and the leg.[7,14,15,17] The diagnosis of this syndrome is often difficult, and its possibility is usually excluded in subjects. This is because there are only a few validated and standardized diagnostic tests to confirm the development of piriformis syndrome. Due to multiple etiologies underlying sciatic nerve entrapment in the subgluteal space such as fibrous bands containing blood vessels, gluteal, and hamstring muscles that have no direct association with the piriformis muscle, the name “deep gluteal syndrome (DGS)” has been suggested recently.[12] Therefore, piriformis syndrome can be regarded as a subtype of DGS, implying that not all DGSSs represent piriformis syndrome.[13]

To diagnose the entrapment of sciatic nerve in the deep gluteal space (piriformis syndrome), researchers have strongly suggested to refer to the patient’s medical history and to ascertain the infliction of blunt trauma to the gluteal area, such as a fall.[3,14] The cause of piriformis syndrome is often an exacerbated activity of hip rotators. It may also occur due to prolonged sitting on hard surfaces.[14] However, the onset of this syndrome is sometimes spontaneous. Typically, a patient complains about buttock pain, which occurs with or without ipsilateral radiation to the ipsilateral posterior thigh. In some cases, it may also extend below the knee and the calf, resembling a typical case of sciatica.[14] Buttock pain or sciatica may be exacerbated with the activity of lower extremities, such as hip adduction and internal rotation that stretches the pyriformis muscle.[14] However, the cardinal feature of this syndrome is sitting intolerance.[3,14] Numbness in various sensory distributions on the involved side are common, but motor weakness in a corresponding distribution is rare.[3] During physical examination of patients, the 2 most common and consistent findings were tenderness and palpation of the greater sciatic notch, and the reproduction of the pain with maximum FADIR of the hip.[12,14]

**Variations of the sciatic nerve and its clinical significance**

The fourth and the fifth lumbar nerve roots combine with the first and second sacral nerve roots, forming the common peroneal and tibial nerves. These 2 types of nerves together constitute the sciatic nerve. As shown in **FIGURE 1**, Beaton and Anson first described the 7 anatomical relationships between the sciatic nerve and piriformis muscle in the year 1937.[2] The conventional and the most common relationship was termed as type A, entailing an undivided sciatic nerve that extends well below the piriformis muscle. According to a cadaver study, the type A sciatic nerve anatomy is prevalent in approximately 80%–90% cases.[11] The type B sciatic nerve is the most common variant, with a prevalence of 10%–15%. In these cases, one division of the sciatic nerve passes through the piriformis muscle, and the other division of the sciatic nerve passes below the piriformis muscle. Type C sciatic nerve is the
The second most common variant, with a prevalence of 1%–3%. In this case, one division of the sciatic nerve passes above the piriformis muscle, and the other division of the sciatic nerve passes below the piriformis muscle (FIGURE 1). The remaining variants, type D–F, are quite rare, each with a prevalence of less than 1%.2,11

The sciatic nerve variation was once considered to be associated with the development of piriformis syndrome,1,7,9,10,11,16,18,19 Today, it is thought to be a normal variant with an uncertain clinical significance.7 No difference was found in the prevalence of variant anomaly in the sciatic nerve and piriformis between cadavers and normal population.19 Piriformis syndrome can occur in patients with normal or variant anatomy of sciatic nerve. In general, patients with a variant anatomy of sciatic nerve are mostly asymptomatic.1 In a large-scale, clinico-radiologic study, researchers investigated the correlation between the symptoms of piriformis syndrome and the MRI of the hip. No relationship was found between the variants of sciatic nerve and the occurrence of piriformis syndrome.9 The symptoms of piriformis syndrome were present in 11.3% of variant hips as compared with 9.0% of normal hips (p=0.39).1

Existing reports of piriformis syndrome caused by sciatic nerve variation

We searched for specific reports on the surgical treatment of piriformis syndrome caused by type C sciatic nerve variation. Given the frequency of cases with piriformis syndrome, we expected that a substantial number of reports would describe direct surgical treatment of piriformis syndrome; however, there were only a few reports of actual surgical treatment. In addition, only a few cases of piriformis syndrome were caused by sciatic nerve variation. Moreover, it was difficult to find the reports appropriately describing the form of variants of sciatic nerve. As shown in TABLE 1, a summary of the reports includes descriptions of the surgical treatment of piriformis syndrome, which is caused by sciatic nerve variation.

Several reasons are given for the lack of reports on the surgical treatment of piriformis syndrome, which is caused by sciatic nerve variation. First, sciatic nerve variations may not be associated with the development of piriformis syndrome. Second, piriformis syndrome is known to respond well to conservative treatment, and new noninvasive treatments are also being developed. Third, existing reports only describe surgical treatment of piriformis syndrome. In these reports, authors did not mention specific subtype of anatomical variations in sciatic nerve. Since 2010, piriformis syndrome has been classified as a subtype of DGS. From this point of time, many reports have described how endoscopic decompression was performed on patients with DGS. In most reports with endoscopic decompression of DGS, it was difficult to find a case in which piriformis syndrome was caused by a specific type of sciatic nerve variation.11

In the present report, we have described 2 cases of typical sciatic nerve entrapment, and it was caused type C variation. According to our focused review (TABLE 1), there were no reports of piriformis syndrome, which showed improvement after surgery as a type C sciatic nerve variation was confirmed as a surgical finding. Piriformis syndromes caused by type D and F variations, which are much rarer than type C, have already been reported (TABLE 1). In the type C variation, there is a highly divided sciatic nerve in the pelvic cavity: one division passes above the piriformis muscle and the other division passes below the muscle.3 In the 2 cases of this report, the peroneal divisions passed above the piriformis muscle. This was detected in the preoperative MRI scan, but was confirmed by electromyographic response and dorsiflexion of the foot with intraoperative nerve stimulation during surgery.
Although variations in the piriformis muscle and sciatic nerve are individual constitutional problems, the exact pathophysiology of abrupt occurrence of sciatic nerve entrapment symptoms is still not well known. Considering occurrence of symptoms of piriformis syndrome following nonsignificant trauma to the buttock, inflammation of the piriformis muscle will result in the release of inflammatory mediators such as prostaglandin, histamine, bradykinin, and serotonin.

CONCLUSION

Very few reports of piriformis syndrome have described how it is caused by sciatic nerve entrapment. In these cases, rare type C variation of the sciatic nerve is associated with the piriformis muscle. In particular, this is the first time that the C variation was confirmed during surgery and an improvement in the pain was reported. In the type C variant, the nerve lying above the piriformis muscle was the peroneal division of the sciatic nerve. This variation of the sciatic nerve and the occurrence of piriformis muscle in a greater sciatic notch does not in itself indicate the existence of piriformis syndrome. However, if rare variations such as type C variants are identified in patients with symptoms of piriformis syndrome, such rare variations should be considered for the cause of sciatic nerve entrapment.

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**TABLE 1.** Reports of surgical treatment of piriformis syndrome, which was caused by specially defined sciatic nerve and piriformis variations

| Year | Authors            | Cases | Type of variation described | Remarks                                      |
|------|--------------------|-------|----------------------------|----------------------------------------------|
| 1992 | Hughes et al.      | 1     | One (type B) of 5 patients (others, type A) | Bifid piriformis muscle with constriction of peroneal division |
| 1994 | Sayson et al.      | 1     | Type G                     | A 36-year-old female, sciatic nerve posterior to the piriformis muscle |
| 1994 | Chen               | 1     | Type D                     | A 28-year-old female, sciatic nerve between bipartite piriformis muscle |
| 1999 | Benson and Schutzer | 1   | (type B) of 14 patients    | Posttraumatic piriformis syndrome            |
| 1999 | Ozaki et al.       | 1     | Type G                     | A 22-year-old female, the divided sciatic nerve above the piriformis muscle |
| 2001 | Spinner et al.     | 1     | Undefined type             | 43-year-old female, poor result despite of decompression |
| 2002 | Fishman et al.     | 6     | Six of 43 patients, type not defined | Not specifically described in the variation group |
| 2002 | Indrekvam et al.   | 4     | 4 of 19 patients, type not defined | 3 of 4 variation patients improved           |
| 2004 | Kobbe et al.       | 1     | 1 of 2 patients, type not defined | An abnormal left sciatic nerve with bifurcation over the piriformis muscle (type B/G?) |
| 2006 | Kosukegawa et al.  | 1     | Type D                     | A 57-old-male, bipartite piriformis muscle, improved with decompression |
| 2008 | Pecina et al.      | 3     | 2 type B and 1 type B      | MRI findings correlated with surgical findings. Improved with surgery. |
| 2022 | Current report     | 2     | Type C                     | Two female patients with post-traumatic (1) and failed back surgery (1) improved |

*Classification by Beaton and Anson. Type A is considered normal and types B–G are considered variant anatomy. Refer to Figure 1 for each variation type.*
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