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Pitout JDD, Church DL, Gregson DB, Chow BL, McCracken M, Mulvey M, Laupland KB (2007). Molecular epidemiology of CTXM-producing Escherichia coli in the Calgary Health Region: emergence of CTX-M-15-producing isolates. Antimicrob. Agents Chemother. 51: 1281-1286.

Pelczar JR, Harley JP, Klein DA (1993). Microbiology: Concepts and Applications. McGraw-Hill Inc., New York, pp. 591-603.

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# **Journal of Neuroscience and Behavioral Health**

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# Journal of Neuroscience and Behavioral Health

Case Report

# A case of intrapelvic sciatic nerve schwannoma presenting as piriformis syndrome

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A 36-year-old man presented with lower back and right lower extremity pain for 10 years and right inguinal pain for 1 year. Magnetic resonance imaging showed a right sciatic nerve mass at the right ventral piriformis, causing sciatica and piriformis syndrome. The tumor was totally resected by a posterior approach. The postoperative histopathological diagnosis was schwannoma (World Health Organization grade 1). Immediately after the operation, his preoperative symptoms disappeared completely.

**Key words:** Intrapelvic sciatic nerve schwannoma, sciatica, piriformis syndrome.

#### INTRODUCTION

Schwannoma is a benign encapsulated slow-growing tumor of schwann cells and is very rare in the sciatic nerve (1%) (Omezzine et al., 2009). Intrapelvic schwannomas are extremely rare. They produce symptoms of sciatica, piriformis syndrome or plantar neuropathy (Tan et al., 2010). We describe a sciatic nerve schwannoma under the piriformis that caused intractable sciatica and mimicked piriformis syndrome.

#### CASE REPORT

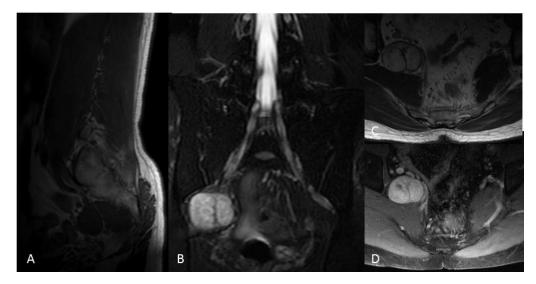
A 36-year-old man presented with a 10-year history of lumbago and right calf and plantar fascia pain. He was diagnosed with an intrapelvic tumor. Conservative treatment had been done for 10 years. He was referred to our hospital because the pain had worsened and spread to his right inquinal region one year before.

Striking his right buttock induced right lower extremity

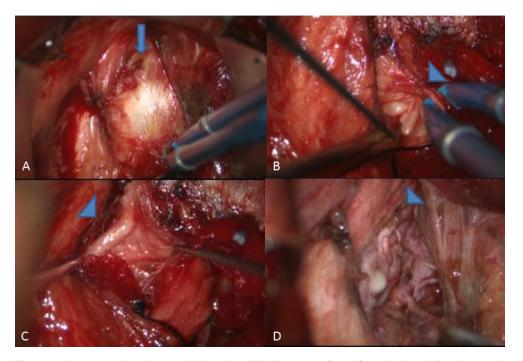
pain. Long periods of standing/sitting and internal and external rotation of his right hip joint increased the pain (Freiberg sign and positive Pace sign). He had slightly dull light touch and temperature senses in his right plantar fascia. Urination time was increased. There was no motor weakness or abnormal deep tendon reflexes in his lower extremities. Magnetic resonance imaging (MRI) showed a  $30 \times 38 \times 32$  mm-size mass along the right sciatic nerve at the right ventral piriformis with low intensity on T1-weighted images, high intensity on T2-weighted images and heterogeneous enhancement with gadolinium (Figure 1).

Under general anesthesia, the patient was placed in a prone position. Nerve integrity was surgically monitored with needle electrodes (Medtronic Inc., Tokyo, Japan) in the right anterior tibialis, the right gastrocnemius and the anal sphincter. A curved skin incision was made on his right buttock. After gluteus maximus cleavage, a small incision was made in the piriformis, revealing the

\*Corresponding author. E-mail: k-ninomiya@nsurg.med.osaka-u.ac.jp. Tel: +81-6-6879-3652. Fax: +81-6-6879-3659.. Author(s) agree that this article remain permanently open access under the terms of the <u>Creative Commons Attribution</u> License 4.0 International License



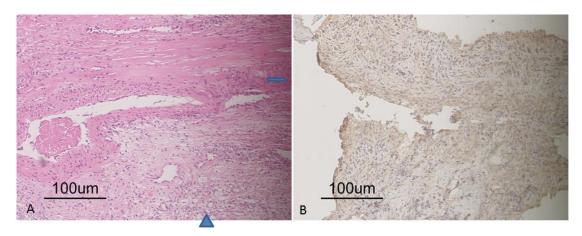
**Figure 1.** Preoperative magnetic resonance imaging of the lumbar spine showing a  $30 \times 38 \times 32$  mm-sized intraextrapelvic mass under the right piriformis muscle (A-D). (A) Sagittal T1-weighted imaging; (B) Coronal T2-weighted imaging; (C) Axial T2-weighted imaging; (D) Axial T1-weighted and contrastenhanced fat-saturation imaging.



**Figure 2.** Intraoperative microscopic imaging. (A) The tumor (arrow) under the piriformis muscle; (B-C) The nerve fascicle with perineurium (arrowhead) was split from the tumor; (D) Operative view after tumor removal.

small incision was made in the piriformis, revealing the superficial part of the tumor. Its rostral and caudal sides were observed and we confirmed that it arose from the sciatic nerve. The tumor was encapsulated with nerve fascicle, showing that active electromyographic responses

responses of the right gastrocnemius were split from the tumor nucleus and preserved (Figure 2). After checking negative electromyographic responses, the rostral and caudal sides of the nerve were cauterized and cut, and almost the entire tumor was resected.



**Figure 3.** A pathological examination of the tumor demonstrated schwannoma, including S100-positive tissue with Antoni A (arrow) and B portion (arrowhead). (A) Hematoxylin and eosin staining and (B) S100 immunohistochemical staining.

Immediately after the operation, all pain and urination difficulty disappeared. There was no motor weakness or other neurological deficits other than numbness of the right buttock and right plantar fascia. The histopathological diagnosis was schwannoma (World Health Organization grade 1) (Figure 3). Postoperative lumbar MRI showed total tumor removal and no recurrence 4 months later.

#### **DISCUSSION**

Sciatic nerve schwannoma is rare, representing only 1% of all schwannomas (Omezzine et al., 2009). It is usually asymptomatic, but tumor mass effects sometimes cause intractable sciatica. Piriformis syndrome accounts for approximately 6 to 8% of sciatica cases (Natsis et al., 2013). This is thought to be caused by sciatic nerve entrapment on the sciatic notch. Generally, it is caused by intrinsic factors, such as fascial pain, anatomical abnormalities, muscular hypertrophy and pyogenic myositis and extrinsic factors, such as hip or pelvic trauma. However, piriformis syndrome can be caused by sciatic nerve perineural cyst (Hwang et al., 2010) or schwannoma (Haspolat et al., 2013), as in this case.

In this case, the slowly growing tumor probably compressed the sciatic and pudendal nerves, thus inducing intractable sciatica, inguinal pain and urination difficulty. Sciatic nerve schwannomas can be purely intrapelvic (Kelso et al., 1993), purely extrapelvic or intraextrapelvic dumbbell-shaped masses (Spinner et al., 2006). Our case was mainly intrapelvic and partially extrapelvic, compressing the sciatic nerve and piriformis, thus causing piriformis syndrome. We chose a posterior approach for resection of this mainly intrapelvic tumor. Several reports have recommended initial tumor capsule incision along the sciatic nerve for en-bloc tumor removal (Russell et al., 2007; Tan et al., 2010). A stimulator probe

first confirmed the motor nerve fascicle with perineurium. Splitting it from the caudal to rostral sides of the tumor, nearly total tumor removal was performed. Only slight numbness of the right buttock and plantar fascia was found postoperatively. We suggest that this operation with intraoperative monitoring and microscopic techniques is equivalent to enucleation. In addition, we should remember that intrapelvic sciatic nerve schwannomas may cause sciatica and piriformis syndrome.

#### Conclusion

We presented a case of intrapelvic sciatic schwannoma which caused sciatica and piriformis syndrome. After total resection of it, his preoperative symptoms disappeared with no major neurological deficit.

#### **Conflict of Interests**

The author(s) have not declared any conflict of interests.

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