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# Plexiform schwannomas of the sciatic nerve:a case report and review of the literature

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#### ABSTRACT

*Background:* Schwannomas grow slowly, mainly in the head and spine. The extremities schwannomas are rare and easily missed, particularly in patients who also have lumbar disc herniation in addition to sciatic schwannomas. We present a unique case of sciatic schwannoma, which has been considered as a lumbar disease in the past until an MRI of the thigh.

Case presentation: A 43-year-old female complained of pain in her low back and left thigh for 10 years. Physical examination showed that her left thigh was swollen and positive Tinel sign. On MRI, we found a series of tumors suspected of schwannomas at the back of her left thigh. After obtaining the patient's consent, we performed intracapsular excision of her tumors. Histological examination of the tumors were consistent with plexiform schwannomas. The patient recovered well after operation and there was no sign of nerve injury or recurrence after follow-up for 11 months. We searched the Pubmed database and found 31 published reports about sciatic schwannomas.

Conclusions: Sciatic schwannomas usually occur in middle-aged women, and the main symptom is pain. In addition to palpation, we should pay attention to Tinel sign during physical examination. MRI is very helpful for diagnosis, but histological examination is the only way to make a final diagnosis. Intracapsular resection is the best method for the treatment of schwannomas, although there is still the possibility of recurrence after operation.

# 1. Introduction

Schwannomas originate from Schwann cells and accounts for 5% of all soft tissue tumors [1]. More than half of schwannomas occur in the head and spine, and they rarely occur in the extremities, especially in the thighs [2]. Schwannomas grow slowly, so there are no obvious symptoms in the early stage especially when they occur in highly muscular areas where they are less likely to be detected [3]. Once schwannomas grow to the point of oppressing the nerve, it can cause pain or paralysis [4]. Coincidentally, if lumbar disc herniation and sciatic schwannomas occur simultaneously, the latter is more difficult to detect because the thigh pain is usually attributed to lumbar disc herniation. Pain medications for lumbar disc herniation may mask the symptoms of schwannomas, ultimately delaying a correct diagnosis. This paper reports a rare case of sciatic schwannoma with lumbar disc herniation, which took 10 years from the onset

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# **Abbreviations**

MRI magnetic resonance imaging.

of symptoms to the final diagnosis.

#### 2. Case Presentation

A 43-year-old woman presented to our hospital with low back and left thigh pain for 10 years. She was previously diagnosed with lumbar disc herniation and denied any family history of inherited diseases. The diagnosis was based on her lower back pain, radiating pain in the left thigh sciatic nerve distribution, positive Lasegue's sign, and MRI showing a herniated disc in the lumbar region. Over the past 10 years, she had been treated with symptomatic therapy for lumbar disc herniation, including oral Celecoxib (100mg bid), acupuncture, and infrared irradiation therapy. Although her lower back and leg pain symptoms were relieved after treatment, she still experienced periodic throbbing pain and weakness in her left leg. Due to the persistent symptoms, her physician recommended referral to a higher-level hospital for surgical treatment, so she presented to our hospital in November 2021. After a series of detailed physical examinations, the results showed that the superficial sensation in the lateral and posterior aspects of her left thigh was reduced compared to the right side, her left quadriceps muscle strength was at a grade 4 level, and her left Lasegue's sign and Tinel's sign were both positive. During the examination, we also found that her left thigh was 8 cm thicker than her right thigh. Based on the severity of her leg pain compared to her lower back pain, we suspected that her pain was not solely due to lumbar disc herniation. Therefore, we performed an MRI with enhanced imaging of her left thigh, which revealed multiple small nodules deep in the posterior aspect of her

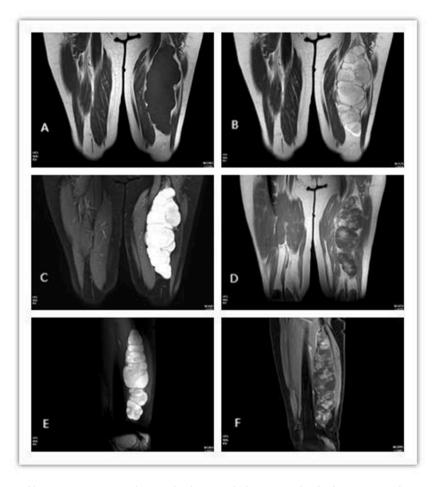


Fig. 1. The tumors presented hypointensity on coronal T1 weighted image, which wpresented as fusiform masses with tapered ends. (A) The tumors presented isointensity on coronal T2 weighted image. (B) The tumors presented hyperintensity on fat suppression image. (C) Coronal contrast-enhanced images presented heterogeneous enhancement. (D) On sagittal view, the tumors were distributed longitudinally along the sciatic nerve. (E, F).

left thigh (Fig. 1). In MRI, the nodules presented as fusiform masses with tapered ends, with low to moderate signal intensity on T1-weighted images and high signal intensity on T2-weighted images. Based on the MRI results and literature review, we highly suspected the possibility of schwannomas.

We recommended surgery to the patient and informed her of the potential risks, such as damage to the sciatic nerve and the possibility of recurrence after surgery. The patient ultimately agreed to undergo the surgery. After administering general anesthesia, the patient was placed in a prone position. The lead surgeon made a 15cm longitudinal incision on the back of the left thigh. After fully exposing the tumor, the surgeon found that the tumor was growing on one side of the sciatic nerve, with a complete and clear boundary (Fig. 2). The surgeon noticed that there were no nerves passing through the tumor, indicating that it was most likely schwannomas rather than neurofibromas. As the capsule of the schwannomas were also part of the nerve tissue, it needed to be preserved. Therefore, while preserving the integrity of the sciatic nerve, we attempted to separate the schwannomas from it. In the end, we removed 21 tumors, with the largest having a diameter of 5cm and the smallest 1.5cm (Fig. 3), Histological examination of the tumors was consistent with schwannomas (Fig. 4). Immunohistochemistry results: S-100(+), SOX10(+), P53(20%+), Ki67(10%+), CD34(+), EMA (-), CD68(-), SMA(-), Desmin(-), CK(-). The patient reported slight pain at the incision site on the second day after surgery, but the previous radiation pain in the left thigh had completely disappeared. On the fourth day postoperatively, the patient reported complete absence of all pain, Physical examination showed negative Lasegue's sign and Tinel's sign, and the sensory disorder in the left thigh disappeared. The patient was subsequently discharged. At 11 months post-discharge, during a telephone follow-up, the patient reported no discomfort symptoms and agreed to publicly share her case. Every procedure carried out during this study complied with the Declaration of Helsinki (as revised in 2013). This study was approved by the ethics committee of the Guangdong Provincial Hospital of Chinese Medicine.

We searched the pubmed database using the keyword [(schwannoma\*) AND (Sciatic)] from January 1, 2001 to December 1, 2022. Finally, 31 case reports were selected, with a total of 33 patients diagnosed with sciatic schwannoma [4–34] (Table 1). There were 13 males and 20 females, with an average age of 45.3 years.

#### 3. Discussion

From Table 1, some patterns of schwannomas can be summarized. Previous studies have found no correlation between gender and the incidence of schwannomas [1]. However, according to our statistics, females seem to be more susceptible to sciatic schwannomas, especially middle-aged women. Obviously, the diagnosis of sciatic schwannomas requires a long time, with an average of 33 months. We believe this is because schwannomas grow very slowly. However, we found that the time required for diagnosis is decreasing year by year, perhaps due to the popularity of MRI. Pain is the primary reason for medical consultation, with only two of the 33 patients having no pain symptoms. More than half of the sciatic schwannomas can be detected through physical examination during the first visit. When the sciatic schwannomas cannot be detected by physical examination, a positive Tinel's sign is an important diagnostic indicator. In terms of tumor morphology, most schwannomas grow in isolation, with few having more than three [5,10,32]. Sciatic schwannomas are usually small, with an average diameter of 5.29 cm. The possibility of schwannomas deterioration is extremely low; so far, only one reported case has shown a sciatic schwannoma transforming into a malignant tumor [10]. Next, we will discuss the above patterns discovered in the literature review.

First of all, why does it take so long from the appearance of symptoms to the correct diagnosis of sciatic schwannomas? The time-consuming diagnosis illustrates the concealment of sciatic schwannomas. We believe that this is due to the slow growth rate of the tumor, and its small size is difficult to detect by palpation, especially when it occurs in the thick thigh. The patient we reported on took 10 years to get the final correct diagnosis, which is obviously much longer than previous reports. We believe that, in addition to the slow growth of the tumor, she also had a lumbar disc herniation. In the past 10 years, the Celecoxib she took had an analgesic effect, which delayed her referral to a higher-level hospital. Compared with the low incidence of sciatic nerve schwannoma, the incidence of

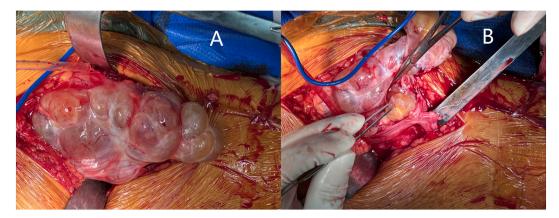


Fig. 2. During the operation, it was found that the tumor grew in clusters and nodules along the sciatic nerve, with a complete capsule and a clear boundary.(A) we separated the schwannomas from the sciatic nerve while preserving the integrity of the capsule.(B).



Fig. 3. The tumors after separation.

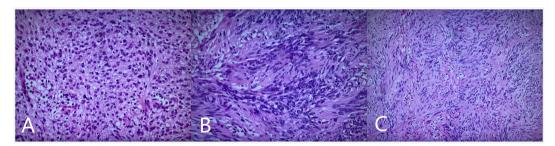


Fig. 4. Histological examination: Alternating arrangement of AntoniA region rich in spindle cells. (A) Verocay bodies: Two rows of closely spaced spindle cells arranged in parallel. (B) Immunohistochemistry showed diffuse positive immunostaining for S-100 protein. (C).

lumbar disc herniation is higher and more well-known. Therefore, when patients with sciatic schwannomas present with pain as the primary symptom, doctors usually first consider sciatic nerve pain caused by lumbar disc herniation. Although MRI has a very high detection rate for tumors, due to the above reasons and economic costs, it is almost impossible to perform an MRI examination on the thigh in the early stages. In summary, we suggest that although the incidence of sciatic schwannomas is low, it should also be taken seriously by doctors and carefully examined. Among all physical examinations, Tinel sign is the most important method of examination, which has important value in revealing this disease.

MRI is one of the most powerful means to make a definite diagnosis. In MRI, schwannomas present as fusiform masses with tapered ends, with low to moderate signal intensity on T1-weighted images and high signal intensity on T2-weighted images [35]. In our case, MRI showed that the tumor consists of a string of well-defined small lumps, resembling a cluster of grapes. This type of multi-nodular growth of schwannomas, which are connected to one another, are called plexiform neurofibroma and accounts for only 5% of all schwannomas [36]. Plexiform schwannomas typically comprise a complete surface capsule and several mucinous nodules with clear borders, usually occur in the skin, and rarely in deep nerves [37,38]. When plexiform schwannomas occur deep in the body, the unique form raises concerns about malignant tumors. However, the vast majority of plexiform schwannomas do not carry a risk of malignant transformation, with only one reported case in the past 20 years [36].

Histological examination is the gold standard for the diagnosis of tumors. Under electron microscope, plexiform schwannomas are usually composed of alternating arrangement of AntoniA region rich in spindle cells and AntoniB region with loose mucoid stroma [37, 38]. Schwannomas are diffusely positive with S-100 protein immunostain, which is helpful to distinguish plexiform schwannomas from malignant tumors [37]. At present, histological examination and immunostaining techniques have been widely used. The limitation of our treatment is the lack of histological examination before surgery. We suggest that all patients with schwannomas diagnosed by MRI should undergo post-puncture histological examination before surgery, although few have done so in the past.

Surgical resection is the best method to treat schwannomas [38]. The available surgical methods include intracapsular excision and extracapsular excision. Since the envelopes of schwannoma are part of nerve, Date found that preserving the envelopes maximized the preservation of nerve function [39]. Some patients have neurological symptoms in the early postoperative period, but most will restore within 1 year [40,41]. It should be noted that plexiform schwannomas have the possibility of postoperative recurrence due to their multinodular growth pattern [38]. The patient in our report showed no signs of tumor recurrence after discharge.

Table 1
Literature review of sciatic schwannomas reported from 2001 to 2022.

Author	Patient information		Symptom				Tinel sign	Tumor morphology	
	Gender	Age	Duration (months)	Pain	Paraesthesia	palpable mass		Number	Diameter (cm)
Yamamoto [5]	F	52	48	Y	N	Y	NR	15	3.5
Maini [6]	F	21	24	Y	N	Y	Y	1	7.6 × 5.4
Rekha [7]	M	60	120	N	N	Y	NR	1	15  imes 10
Consales [8]	F	39	NR	Y	N	N	NR	1	5.5 × 4.5
Kralick [9]	M	58	5	Y	N	N	N	1	3.5
Lee [10]	M	73	240	Y	N	Y	NR	NR	$30 \times 5 \times 5$
Blanchard [11]	F	42	84	Y	Y	N	N	1	8.5
Hamdi [12]	M	45	3	Y	N	Y	N	1	7.8
Omezzine [13]	F	42	12	Y	Y	Y	NR	1	5
Tan [14]	M	51	12	Y	N	N	NR	1	NR
Rhanim [4]	M	37	36	Y	N	Y	_	1	3
Haspolat [15]	M	60	NR	Y	Y	N	NR	1	NR
Eroglu [16]	F	40	12	Y	N	Y	+	1	6
Chahbouni [17]	F	48	24	Y	Y	Y	NR	1	4
	F	52	36	Y	Y	Y	NR	1	5
Mansukhani [18]	F	46	48	Y	N	N	+	1	$2.5 \times 2$
Chikkanna [19]	M	40	NR	Y	N	Y	_	2	5 & 1
Godkin [20]	F	40	8	Y	N	Y	NR	1	9 × 6
Mezian [21]	F	27	48	Y	N	N	+	1	4 × 3
Munakomi [22]	F	69	48	Y	N	N	NR	1	3
As-Sultany [23]	F	39	6	N	N	Y	NR	2	$4.5 \times 6 \times 12\&3 \times 3.5 \times 6$
Naik [24]	M	34	12	Y	N	N	+	1	$5 \times 4 \times 3$
Guedes [25]	F	40	7	Y	N	Y	+	1	NR
Chagou [26]	M	30	12	Y	N	Y	+	1	2
Maes [27]	F	50	NR	Y	N	Y	_	1	$7 \times 4.4 \times 3.3$
Telera [28]	F	47	12	Y	N	N	+	1	3
	F	79	12	Y	N	N	+	1	$7 \times 5 \times 4$
Wu [29]	M	27	24	Y	N	N	+	1	$2 \times 1.7 \times 2.2$
George [30]	F	57	12	Y	N	N	+	1	3
Utomo [31]	F	35	8	Y	N	N	NR	1	$13.9 \times 1.91$
Erdoğan [32]	M	22	14	Y	Y	N	+	7	3
Shariatzadeh [33]	M	45	NR	Y	N	Y	_	4	4
Zhang [34]	F	49	2	Y	Y	Y	NR	1	3  imes 2  imes 2

 $Abbreviations: M = male, F = female, Y = yes, N = no, NR = no \ report. \\$ 

#### 4. Conclusions

In summary, we report a rare case of the largest sciatic plexiform schwannomas to date. By reviewing the past literature, we identified certain characteristics of sciatic schwannomas, including a higher incidence in middle-aged women, concealment and difficulty in differentiation from lumbar spine diseases. In terms of diagnosis, Tinel's sign can assist in early detection, MRI can confirm tumor morphology, and histological examination can clarify tumor nature. As for treatment, intracapsular excision is the best method. Additionally, doctors should have detailed communication with patients before surgery, including risks of nerve damage, the necessity of postoperative histological examination, and the possibility of postoperative recurrence.

#### Author contribution statement

All authors listed have significantly contributed to the investigation, development and writing of this article.

# Data availability statement

Data will be made available on request.

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### Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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