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

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Case Report

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Abstract

Background: Plexiform sciatic schwannoma is a rare tumor. Schwannoma is deceptive when it appears on the sciatic nerve. We report a rare case of plexiform sciatic schwannoma that was initially diagnosed as a lumbar disease.

Case Presentation: A 43-year-old female complained of pain in her low back and left thigh for 10 years. Physical examination and MRI confirmed that she had a rare plexiform sciatic schwannoma. After obtaining the patient's consent, we performed intracapsular excision of her tumor. The patient recovered well after operation and there was no sign of nerve injury.

Conclusions: Schwannomas in the sciatic nerve are misleading and doctors should pay attention to physical examination and MRI. Intracapsular excision is the best treatment for schwannomas.

Background

Schwannoma originates 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 extremity schwannomas are more common in the upper extremities\(^2\). Schwannoma grows slowly and has limitations, so there are no obvious symptoms in the early stage. Especially if schwannoma occurs in muscle-rich areas, it is not easy to detect\(^3\). When the schwannoma grows to the point of oppressing the nerve, the patient will have symptoms such as pain and paralysis. Therefore, only when the patient has symptoms of nerve compression, the schwannoma hidden in the muscle has a chance to be found\(^4\). Coincidentally, if a patient has both lumbar disc herniation and sciatic schwannoma, the former will make the latter more difficult to detect because the thigh pain are often blamed on lumbar disc herniation.

Case Presentation

A 43-year-old woman came to our hospital with low back and left thigh pain for 10 years. Physical examination showed lumbar tenderness and positive Lasegue’s test. Lumbar MRI indicated lumbar disc herniation. According to the above results, the previous diagnosis was lumbar disc herniation. Over the past 10 years, her low back pain has been relieved with conservative treatment. However, the pain in her left thigh is getting worse. In November 2021, she came to our hospital again because of pain in her left thigh. After a series of more detailed physical examinations, we found that her left thigh was 8 cm thicker than her right thigh and her left thigh tinel sign(+). We performed an enhanced MRI scan of her thigh and found many small lumps deep in the back of the left thigh (Fig. 1). In MRI, the lumps 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. According to MRI, they are very similar to schwannomas.

We recommend surgery and inform patients that the operation may damage the sciatic nerve. The patient was placed in the prone position after being general anesthesia. We cut the skin and subcutaneous tissue longitudinally along the back of the thigh to reveal complete tumors. The tumors were connected to the sciatic nerve with complete capsules and clear boundary (Fig. 2). The capsule of schwannoma is part of the nerve, so it should be retained. While preserving the intact nerve, we tried to isolate the schwannoma thoroughly. Eventually, twenty-one tumors were removed, the largest of which was 5 cm in diameter and the smallest was 1.5 cm in diameter (Fig. 3). Histological examination of the tumors were consistent with schwannomas (Fig. 4). Immunohistochemistry results: S-100(+), SOX10(+), P53(20%+), Ki67(10%+), CD34(+), EMA(-), CD68(-), SMA(-), Desmin(-), CK(-). The patient recovered well after operation, and the symptoms of thigh pain disappeared obviously. 10-month follow-up showed no signs of nerve injury. After communication, she has agreed to publish her case.

Discussion

We searched the pubmed database using the keywords “schwannoma” and “sciatic nerve” from 2012 to 2022. Finally, 21 case reports were selected, with a total of 23 patients diagnosed with sciatic schwannoma\(^4\text{-24}\) (Table 1). There were 8 males and 15
females, with an average age of 44 years. Among the 23 patients, 22 patients complained of pain, and 5 of them had paraesthesia at the same time. The average duration of symptoms in these patients was 20.05 months before diagnosis. Only half of the patients’ tumors can be detected by palpation. Of the 11 patients whose tumors were not detected on the body surface, 8 were positive for tinel sign, and the rest were unknown. Only 4 patients had multiple schwannomas. The largest tumor was 13.9 cm in length and the smallest was 1 cm, with an average diameter of 4.27 cm.

Table 1 Literature review of sciatic schwannomas reported from 2012 to 2022

<table>
<thead>
<tr>
<th>Author</th>
<th>Patient information</th>
<th>Symptom</th>
<th>Tinel sign</th>
<th>Tumor morphology</th>
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<td>Pain</td>
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We report a rare case of plexiform neurilemmoma in the sciatic nerve, which has only been reported once before\textsuperscript{22}. Erdoğan found 7 schwannomas on the left thigh of a 22-year-old man, whose morphological and histological examination were similar to those in this case\textsuperscript{22}. The young man, like the woman in this case, received treatment for the lumbar spine for more than a year before correct diagnosis. Previous studies have shown that there is no difference in the incidence of schwannomas.
between men and women\textsuperscript{[1,25]}. However, our statistics show that women are more likely to develop sciatic schwannomas. Patients with sciatic schwannoma mainly take pain as the first symptom, and sometimes have sensory disorders. Schwannomas are usually small and solitary, so when located in the thigh, the detection rate of palpation is low\textsuperscript{[25]}. We believe that schwannoma on the sciatic nerve is obviously misleading. For patients who complain of pain in the sciatic innervation area, doctors tend to consider the possibility of lumbar disease. Therefore, it usually takes more than 1 year from the onset of symptoms to the correct diagnosis. Although the incidence of sciatic schwannoma is very low, it should also attract the attention of doctors, and careful physical examination is necessary. When schwannoma cannot be found by palpation, Tinel sign and MRI are important examination methods. The Tinel sign of schwannoma is usually positive.

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\textsuperscript{[25]}. Plexiform schwannoma is a rare subtype of schwannoma, contains multiple well-defined myxoid nodules with a complete capsule on the surface\textsuperscript{[26]}. The diagnosis of plexiform schwannoma depends more on the exploration of tumor morphology during operation. Complete capsule and clear boundary are the main features of benign schwannoma. Plexiform schwannoma usually occurs in the skin and rarely in the major nerves\textsuperscript{[27]}. When it occurs deep in the body, the astonishing degree of mitosis of plexiform schwannomas will raise concerns about malignancy. However, the vast majority of plexiform schwannomas are not at risk of malignant\textsuperscript{[26]}.

Histological examination has always been regarded as the gold standard for the diagnosis of tumors. Under electron microscope, plexiform schwannomas are usually composed of Antonia type An and well-structured Verocay bodies\textsuperscript{[26,27]}. Schwannomas are diffusely positive with S-100 protein immunostain, which is helpful to distinguish plexiform schwannomas from malignant nerve sheath tumors\textsuperscript{[26]}.

Surgical resection is the best way to treat schwannoma with obvious clinical symptoms\textsuperscript{[27]}. Because the capsule of schwannoma is a part of nerve, Date found that intracapsular excision of schwannoma with reserved capsule can preserve nerve function to the maximum extent, which is better than extracapsular excision\textsuperscript{[29]}. Some patients have neurological symptoms in the early postoperative period, but most will restore after 1 year\textsuperscript{[30]}. It should be noted that plexiform schwannomas have the possibility of postoperative recurrence because of multinodular growth pattern\textsuperscript{[27]}. The case was followed up for 10 months without recurrence.

**Conclusions**

In summary, we report a rare case of the largest plexiform sciatic schwannoma to date. Literature review suggests the following 5 points:

1. The incidence of sciatic schwannoma in females is higher than that in males.
2. Schwannoma in the sciatic nerve is misleading and its diagnosis is often delayed for more than one year.
3. Detailed physical examination and MRI are helpful to find neurilemmoma. In particular, the positive Tinel sign is of great significance to the unpalpable tumors.
4. Plexiform sciatic schwannoma is a rare tumor and needs to be distinguished from malignant tumor. It can be distinguished by intraoperative exploration and histological examination.
5. Intracapsular excision is the best treatment without sequelae, but there is a possibility of recurrence. All these need to be communicated to the patient before operation.

**Declarations**

Consent for publication: The patient has provided informed consent for publication of the case.
Ethical Approval

The publication of this case report has been approved by the Ethics Committee and the informed consent of the patient.

Competing interests

The authors deny that they have competing interests.

Authors' contributions

Yan J and Zhou R wrote the main manuscript text. Cao X was the chief surgeon of the operation and revised the manuscript. Yan J and Liu B assisted the operation and prepared the photographs. All authors reviewed the manuscript.

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Availability of data and materials

The original data supporting the conclusions of this article will be provided by the authors.

References


**Figures**
Figure 1

The tumors are hypointense on T1-weighted images while hyperintense on T2-weighted images and contrast enhancement at the peripheral edges shown after contrast.

Figure 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.
Figure 3

The tumors after separation

Figure 4
Histological examination: consistent with schwannoma