

## Pediatric sciatic neuropathy: clinical presentation and long term follow up

### Neuropatía ciática en pediatría: presentación clínica y seguimiento a largo plazo

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#### What do we know about the subject matter of this study?

Sciatic neuropathy in children is an uncommon condition, which most frequent causes are due to trauma and compression. In low-income countries, intramuscular injections are the leading cause of this entity and, in addition, there is a lack of information on its long-term prognosis.

#### What does this study contribute to what is already known?

In this study, we observed that trauma and compression were the most frequent causes of sciatic neuropathy and there were no cases due to intramuscular injection. In addition, we found that in the long term motor sequelae frequently appeared, as well as orthopedic complications, requiring surgery to correct them.

#### Abstract

Sciatic neuropathy is rare and difficult to diagnose in pediatrics, and its long-term course has not been completely understood. **Objective:** To analyze the clinical presentation and evolution of a group of pediatric patients with sciatic neuropathy. **Patients and Method:** Retrospective analysis of the clinical characteristics of pediatric patients with sciatic neuropathy treated in two hospitals of Santiago between 2014 and 2018. Locomotor examination, muscle trophism, deep tendon reflexes, gait, sensation, and pain were assessed. Sciatic nerve conduction study and electromyography (EMG) were performed, and magnetic resonance imaging (MRI) in three patients. **Results:** Six patients were included with an average age of 11.8 years. The etiologies

#### Keywords:

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were traumatic (N = 2), by compression (N = 2), vascular (N = 1), and tumor (N = 1). All of the 6 patients presented foot drop and Achilles tendon hyporeflexia/areflexia, and 5 patients presented severe neuropathic pain. The EMG showed involvement of the sciatic nerve rami and dependent muscles. In two patients, a pelvic girdle and lower limbs MRI was performed, showing selective muscle involvement in sciatic territory. One patient underwent a lumbosacral plexus MRI, and subsequently histological study showing a benign neural tumor. Out of the three patients who were followed-up longer than one year presented motor sequelae and gait disorder. **Conclusion:** Sciatic neuropathy in the study group was secondary to different causes, predominantly traumatic and compressive etiologies. The three patients that were in a long-term follow-up presented significant motor sequelae. In most of the cases, neural injury was associated with preventable causes, such as accidents and positioning in unconscious children, which is crucial in the prevention of a pathology with a high sequelae degree.

## Introduction

Sciatic neuropathy in pediatric age is a rare entity in our sphere<sup>1</sup>. Some series show that between 5 and 6% of all electrophysiological studies (EPS) performed on pediatric patients were mononeuropathies<sup>2</sup>, and only 25% of them involve the sciatic nerve. In adulthood, the so-called 'sciatica' is a different condition, which often corresponds to degenerative disc disease, presenting a significantly higher prevalence<sup>3</sup>. In pediatric patients, sciatic neuropathy may be unilateral or bilateral and appears with motor and sensory deficits in the affected lower limb, often associated with difficult-to-control neuropathic pain. It has different causes, such as due to trauma or compression, vascular, tumor, post-viral and idiopathic ones<sup>1</sup>. Historically, the sciatic neuropathy study was based on electrophysiology, however, neuroimaging has been recently incorporated as a new diagnostic tool, although it is still seldom used. The available literature is usually limited to case reports, with few large pediatric series of sciatic neuropathy published to date<sup>1,4</sup>.

Long-term follow-up has been rarely reported in children with sciatic neuropathy<sup>5</sup>. The few reports indicate that its prognosis depends on the initial severity of the lesion, its etiology, and management. Causes due to trauma or compression are associated with a better prognosis than tumors and vascular ones<sup>1</sup>. Regarding its management, the intervention through early neurolysis is important, since in post-injection neuropathies improves the long-term prognosis<sup>5</sup>. The sequelae observed include decreased longitudinal growth of the affected lower limb, muscle atrophy, and distal paresis<sup>5</sup>.

The objective of this series is to describe a group of children with sciatic neuropathy of diverse etiology, from its presentation to its long-term course and describing the diagnostic study, as well as the general management of this entity. We especially detail the imaging findings and their contribution to the diagnosis and follow-up of our patients.

## Patients and Method

This study was approved by the local Ethics Committees (South East Metropolitan Health Service and Faculty of Medicine of Pontifical Catholic University of Chile).

### Clinical assessment

Between January 2014 and December 2018, a clinical characterization was carried out of pediatric patients with sciatic neuropathy seen at the Dr. Sótero del Río Hospital and the UC CHRISTUS Clinical Hospital, by reviewing clinical records and complementary studies. Also, personal and family history was recorded as well as the cause of the lesion. Motor skills were assessed including the strength of muscle groups (classify according to the Medical Research Council scale<sup>6</sup>), muscle atrophy, presence and quality of deep tendon reflexes, and gait. The sensory function and presence of pain and its management were assessed. In cases where there was more than a two-year follow-up, the treatment administered and the evolution were described.

### Electrophysiological study (EPS)

The EPS was performed by pediatric neurologists using the Sierra® Summit™ equipment (Cadwell, Kennewick, United States) and the Sierra® Wave™ equipment (Cadwell, Seattle, United States), assessing the nerve conduction of the sciatic nerve branches and subdivisions (superficial peroneal sensory and sural nerve conduction, and peroneal and tibial motor nerve conduction). Electromyography (EMG) was performed of muscles innervated by sciatic, peroneal, and tibial nerves with needle electrodes.

### Magnetic Resonance Imaging (MRI)

Three patients underwent MRI of the pelvic girdle and lower limbs. The study was carried out using the Philips Achieva 1.5-T MRI system (software Release,

Philips Medical Systems, The Netherlands) and the MAGNETOM® Avanto 1.5T MRI system (Siemens, Erlangen, Germany).

The MRI protocol for muscle evaluation consists of axial and coronal T1 and coronal STIR, modified from what was previously published<sup>7</sup>. The modified Mercuri scale was used to assess muscle involvement. This scale allows classifying the involvement degree into four groups according to the hypersignal percentage in each muscle (normal, mild, moderate, severe)<sup>7,8</sup>.

The lumbosacral plexus sequence includes a coronal 3D STIR (Short-Tau Inversion Recovery; TE = 254 ms, TR = 2,700 ms, TI = 160 ms, FOV 350 mm, voxel size 1.1 x 1.1 x 1.1 mm; acquisition time 4:38 min) and axial T2 with mDIXON fat saturation (TE = 73 ms, TR = 6890 ms, FOV 300 mm, resolution 0.9 x 0.9 mm in plane, cut thickness 4.5 mm, acquisition time 3:49 min). The use of contrast was upon the radiologist's discretion.

## Results

### Demographic distribution and etiology

Six female patients were included aged between 3 and 14.9 years (average 11.8 years), presenting different etiologies (Table 1).

Among these etiologies, two girls presented trauma causes, the first one (Patient 1) due to a stab wound in the right gluteal region and the second one (Patient 2) due to a left hip dislocation resulting from a motor vehicle accident. There were two patients who presented compressive etiology, the first one (Patient 3) due to prolonged supine position during hospitalization with

a pharmacologically altered state of consciousness, and the second one (Patient 4) due to endoscopic transsphenoidal surgical positioning. One patient (Patient 5) presented Hemolytic Uremic Syndrome that required prolonged hospitalization. During the course of the syndrome, the patient was alert, not under sedation or paralysis, she presented with acute neuropathy, which was considered of vascular origin. The last patient (Patient 6) presented a tumor-caused neuropathy due to a hybrid schwannoma/perineurioma (Table 1).

### Clinical characteristics

All patients presented foot drop secondary to tibialis anterior muscle paresis (peroneal branch of the sciatic nerve). Also, different degree of difficulty was observed in foot eversion due to peroneus muscle paresis (peroneal branch of the sciatic nerve), and in foot inversion due to tibialis posterior muscle paresis (tibial branch of the sciatic nerve). It was observed that all patients presented *steppage* gait with no Achilles deep tendon reflexes response in three cases and a diminished one in the remaining three.

Regarding sensory characteristics, five girls (patients 1 to 5) presented hypoesthesia in the superficial peroneal and sural area, in addition to acute neuropathic pain in leg and foot (Table 1).

### Electrophysiological study (EPS)

Sensory nerve conduction showed superficial peroneal nerve involvement in four patients and sural nerve involvement in all patients. Motor nerve conduction showed peroneal nerve involvement in six cases and tibial nerve involvement in five ones.

EMG with needle electrode in the peroneal nerve

**Table 1. Etiology and initial clinical manifestations in 6 girls with sciatic neuropathy**

P	Sex	Age, years	Etiology	Side	Motor manifestations	Sensitive manifestations
1	F	14,9	Traumatic: penetrating gluteal wound	R	Foot dorsiflexion M0, foot eversion M0, foot inversion M2	NP L5-S1, leg and foot hypoesthesia
2	F	9,9	Traumatic: hip luxation	L	Foot dorsiflexion M0, foot eversion M3	NP, foot hypoesthesia
3	F	14,4	Compressive: long supine decubitus	L	Foot dorsiflexion M0	Foot NP, posterior leg hypoesthesia
4	F	9	Compressive: long supine decubitus	L	Foot dorsiflexion M0, plantar flexion M0	NP, foot hypoesthesia
5	F	7	Vascular: HUS	L	Foot dorsiflexion, foot eversion, foot	Foot NP, thigh, leg and foot
6	F	3	Tumor: intraneural Schwannoma/perineurioma	L	Foot dorsiflexion M0, foot eversion M1, foot inversion M3	Late lumbar pain

P: patient. F: female. R: right. L: left. M0: no contraction. M1: weak contraction. M2: active movement without gravity opposition. M3: active movement against gravity force. NP: neuropathic pain. OTR: osteotendinous reflexes. HUS: Hemolytic Uremic Syndrome.

area showed acute denervation and decrease or absence of motor unit recruitment in all patients. Three out of five patients presented acute denervation in the tibial nerve area, with a decrease or absence of motor unit recruitment in all of them. The femoral region of five patients and the upper gluteal region of three patients were evaluated, which were not related to the sciatic region, showing no alterations in the EPS (Table 2).

A follow-up EPS was performed on two patients. In Patient 2, it was carried out five months after the first study showing no changes. In Patient 3, the study was performed two months after the first one, where the EMG showed chronic reinnervation without the acute denervation activity described in the first evaluation.

### Imaging study

Two girls (Patients 1 and 2) underwent muscle MRI that showed selective fatty infiltration and muscle atrophy in sciatic, peroneal and tibial regions of the affected limb. The tibia was more affected than the thigh and the peroneal area more than the tibial one. The quadriceps (femoral nerve) was normal in both cases (Figure 1).

The lumbosacral plexus MRI performed on Patient 6 showed signs of significant increase in thickness and signal in T2 of the L4 and L5 nerve roots, including the dorsal ganglion, extending to the sciatic nerve, without the use of contrast (Figure 2), suggestive of an intraneural perineurioma (formerly hypertrophic localized

neuropathy) versus a chronic inflammatory demyelinating polyneuropathy. A histological study of the sciatic nerve was performed and concluded an intraneural/hybrid schwannoma/perineurioma.

### Initial therapeutic approach

The kinesiology and occupational therapy team treated all cases, using supramalleolar orthosis. All patients required some kind of pharmacological treatment. Five out of six girls received pharmacological treatment due to neuropathic pain. Because of the acute pain, the five affected patients could not fall asleep. The drugs used were GABA analogs (pregabalin) associated with a tricyclic antidepressant (amitriptyline). In all cases, the symptom was completely managed within a few weeks. Before the biopsy, Patient 6 was initially managed as a possible inflammatory neuropathy and was treated with prednisone 2 mg/kg/day for 1 week, without evident response.

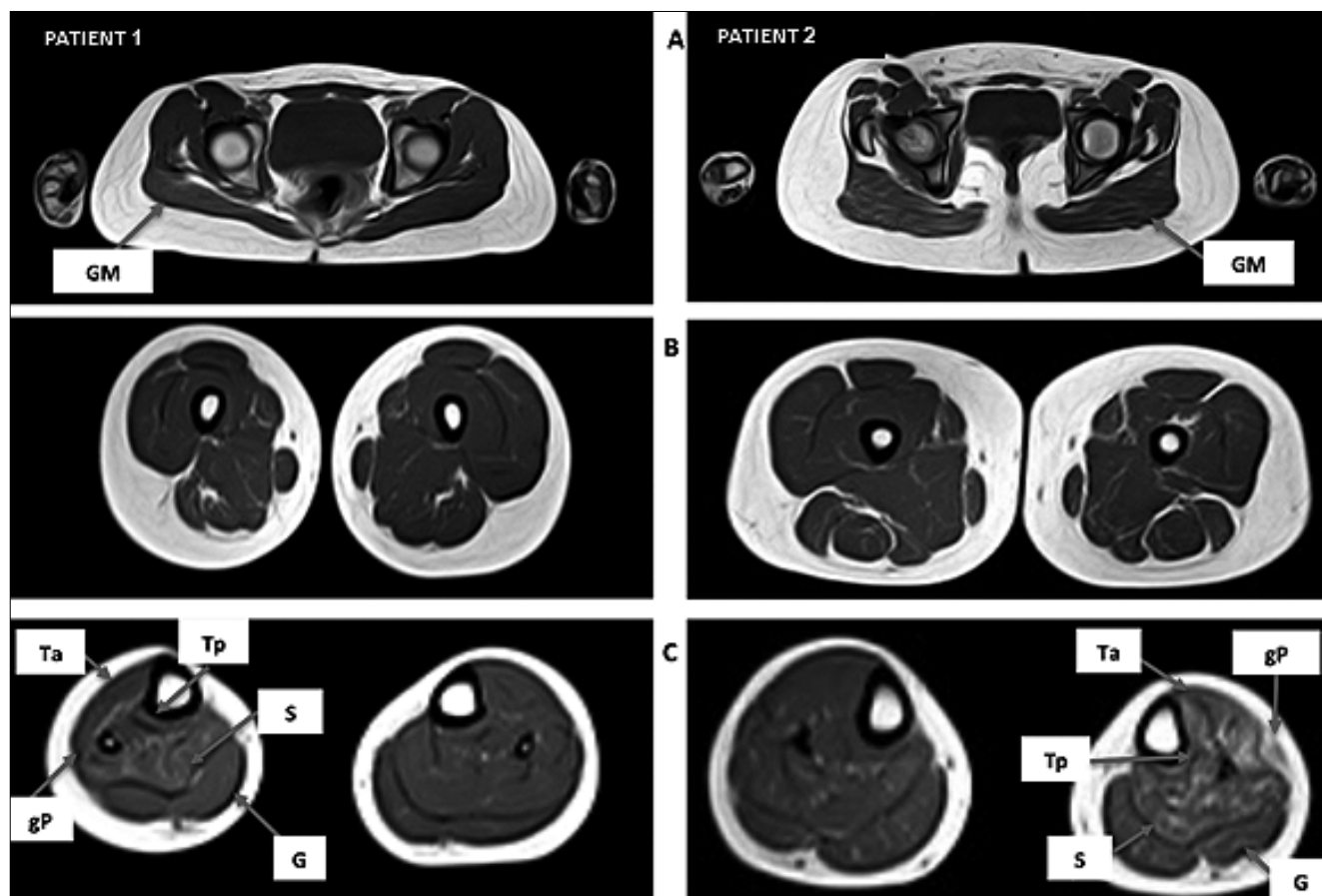
### Clinical follow-up

We were able to clinically follow-up three patients for more than two years. Two cases presented neuropathies due to trauma and one due to a tumor. The first case (Patient 1, 24-months follow-up) had neuropathy due to trauma secondary to stab wound that maintained hypoesthesia in the tibial and peroneal region. She also persisted with a more evident motor involvement in the peroneal area than in tibial one. In addi-

**Table 2. Electromyography features of 6 girls with sciatic neuropathy in affected limb: first evaluation**

Case	Sensitive nerve conduction		Motor nerve conduction		Electromyography	
	Superficial Peroneal	Sural	Peroneal	Tibial	Peroneal territory	Tibial territory
1	UA	Absent	Absent	Absent	Tibialis anterior PSW/F at rest. No MU recruited	Gastrocnemius PSW/F at rest. No MU recruited
2	Absent	Absent	Absent	Absent	Tibialis anterior OA+F en reposo. No recluta UM	Gastrocnemius PSW/F at rest. No MU recruited
3	Absent	Absent	Absent	Decreased amplitude	Tibialis anterior PSW/F at rest. No MU recruited	Gastrocnemius Normal activity at rest. Decreased MU recruitment
4	UA	Absent	Absent	Decreased amplitude	Tibialis anterior Extensor hallucis longus PSW/F at rest. No MU recruited	Gastrocnemius PSW/F at rest. Decreased MU recruitment
5	Absent	Decreased amplitude	Decreased amplitude	Decreased amplitude	Tibialis anterior PSW/F at rest. No MU recruited	Gastrocnemius Normal activity at rest. Decreased MU recruitment
6	Absent	Decreased amplitude	Decreased amplitude	Normal	Tibialis anterior Peroneus longus long head PSW/F at rest. Decreased MU recruitment	Gastrocnemius UA

UA: unavailable. PSW: positive sharp waves. F: fibrillation. MU: motor units.



**Figure 1.** Magnetic resonance images with muscle protocol evaluation of hip girdle and lower extremities of patients 1 and 2. Axial T1 in (A) hip girdle, (B) thighs and (C) leg. Right sciatic neuropathy in patient 1, after 2 months of follow up. There is light involvement (hyper-signal and atrophy) in (A) gluteus maximus and (B) thigh. (C) Moderate involvement of the anterior compartment of the leg, soleus, tibialis posterior and peroneal group. Light involvement of gastrocnemius. Left sciatic neuropathy in patient 2, after 3 years of follow up. (A) Normal gluteus maximus. (B) Light thigh involvement. (C) Leg anterior compartment, soleus and tibialis posterior muscles moderate involvement. Peroneal group severe involvement. Gastrocnemius light involvement. GM: gluteus maximus, Ta: tibialis anterior, Tp Tibialis posterior, S: soleus, gP: peroneal group, G: gastrocnemius.

tion, she did not achieve dorsiflexion or eversion of the foot on her last assessment. The patient partially recovered mobility of innervated muscles by the tibial nerve, achieving movement against gravity but not resistance in plantar flexion and foot inversion. She was referred to an orthopedist and is waiting for tendon transfer surgery. Currently, she requires the use of orthoses for walking.

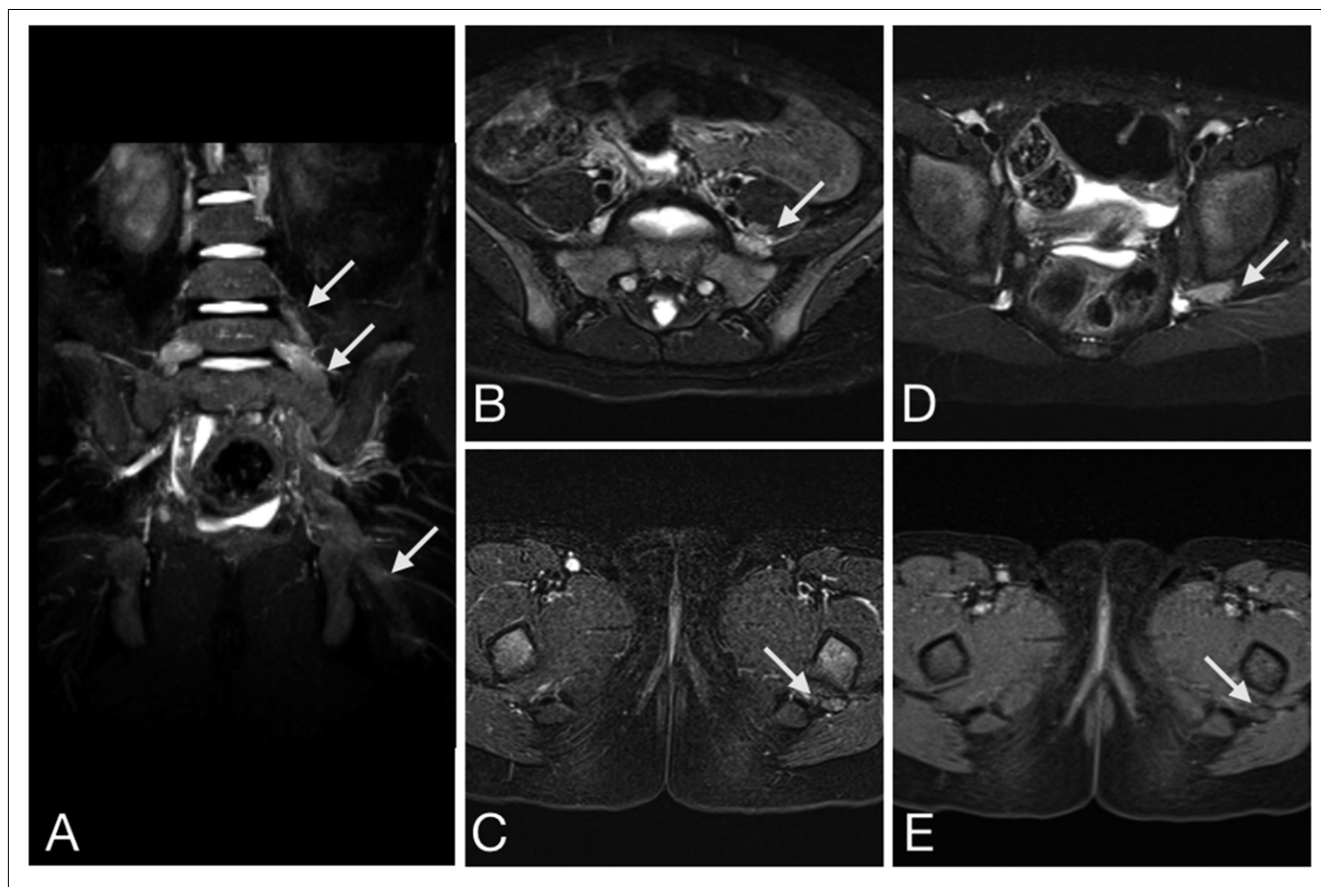
Patient 2 had a 48-months follow-up. She was diagnosed with neuropathy due to trauma secondary to hip dislocation. She regained full foot sensation in the 12th month of follow-up. Regarding motor characteristics, she recovered dorsiflexion in her foot (peroneal branch of the sciatic nerve), achieving movement against gravity and resistance after 48 months of follow-up, but could not achieve inversion (tibial branch of the sciatic nerve). At 36 months of follow-up, she had a corrective

surgery due to valgus heel (osteotomy and talonavicular arthrodesis), with satisfactory results. The patient achieved gait without the use of an orthosis, with slight claudication of the affected limb.

Patient 6 had neuropathy due to a tumor. She maintained the decreased strength observed in the initial assessment, with no evident recovery in two years of follow-up. She evolved with varus foot and was referred to an orthopedist for surgical correction. She presented *steppage* gait requiring orthotics to achieve normal gait.

## Discussion

We reported a series of six pediatric patients with sciatic neuropathy, a rare entity in this population<sup>1,9</sup>. It



**Figure 2.** Lumbar plexus MR images of patient 6, with neurography protocol 7 months after symptoms onset. (A) STIR 3D coronal section with 10 mm width MIP format, (B, C, D) T2 axial weighted and fat saturation with DIXON technique at lumbar spine, hip and superior thigh respectively and (E) T1 axial weighted, with fat saturation DIXON technique and intravenous contrast. The images show marked hyper-signal in T<sub>2</sub> and enlargement of L4 and L5 roots extending caudally and involving the sciatic nerve up to the thigh (arrows). No abnormal reinforcement is observed. No denervation signs in visible muscle groups are observed.

has an uncertain prognosis that, in our series, was associated with motor sequelae and long-term gait disorder in at least half of the cases.

The most frequent causes were due to trauma and compression, which is in line with that described in the literature<sup>1</sup>. Unlike other series<sup>4,10</sup>, in ours, there were no cases of sciatic neuropathy secondary to intramuscular injection. In this series, two patients presented sciatic neuropathy due to trauma (hip dislocation and buttocks stab wound). It has been reported that occasionally, hip pathology and procedures, such as arthroplasty<sup>11</sup> and dislocation<sup>12</sup>, are associated with sciatic neuropathy, due to their anatomical proximity. Likewise, penetrating wounds have already been reported in 2/53 children in a series of pediatric patients with sciatic neuropathy<sup>1</sup>. The compression of the sciatic nerve, as occurred in two of our patients, has also been described in pediatric and adult reports with altered state of consciousness, hospitalized in intensive care unit<sup>13,14</sup>, and in children undergoing different surgeries<sup>1</sup>.

On the one hand, sciatic neuropathy in Hemolytic Uremic Syndrome (Patient 5) has not been previously reported. However, the presence of sciatic neuropathy has been described in other types of vasculitis, such as the post-streptococcal one<sup>1</sup> and, on the other hand, cases of pediatric and adult patients with peroneal neuropathy and optic neuropathy have been described in the Hemolytic Uremic Syndrome. The mechanism proposed is ischemia due to thrombotic microangiopathy involving the vasa nervorum<sup>15,16</sup>.

In the case of Patient 5, a compression mechanism could also be considered, since there was a prolonged hospitalization. However, the onset of symptoms occurred while the patient had already recovered wakefulness and mobility, so a compressive cause seems less likely. In neuropathy due to compression associated with prolonged supine position, especially related to the period of altered state of consciousness and immobilization, pain or neurological deficit is usually evident upon return to wakefulness<sup>1</sup>, a different situation

than that occurred in this patient.

In Patient 6, imaging findings suggest an intraneural perineurioma versus chronic inflammatory demyelinating mononeuropathy/polyneuropathy<sup>17</sup>. The absence of reinforcement on MRI is more suggestive of a chronic inflammatory neuropathy, but in this case, the biopsy concluded that the lesion was an intraneural schwannoma/perineurioma without elements of malignancy, an entity that has not been previously described at the sciatic nerve level<sup>18</sup>.

Intraneural perineurioma, also called localized hypertrophic neuropathy of the sciatic nerve, is also a rare benign tumor and is probably under-diagnosed. Its typical clinical presentation is a mononeuropathy of large nerves (e.g. sciatic nerve) in a young or pediatric patient, with slowly progressive non-painful paresis, and minimal sensory involvement, as observed in our patient. Its management is controversial since some experts recommend observation only, while others suggest surgical excision of the tumor with or without nerve graft or transfer<sup>19,20</sup>. In this case, due to its clinical-radiological similarity and because it is a benign tumor, it was decided to perform expectant management as suggested in intraneural perineurioma.

Regarding clinical manifestations, sensory symptoms (neuropathic pain and paresthesias) were observed in both peroneal and sural areas and the onset of foot drop. However, when muscle strength was assessed in a targeted manner, paresis was more severe in characteristic muscles of the peroneal nerve than in those of the tibial one. This finding could be related to the sciatic nerve anatomy and the higher susceptibility of the portion that forms the peroneal branch<sup>21</sup>.

In relation to the electrophysiological study, all cases presented altered sensory and motor nerve conduction in the peroneal nerve and, less severely, in the tibial one. EMG with needle electrode showed a variable involvement degree, with similar anatomical location as described in nerve conduction. The electrophysiological study is essential in sciatic neuropathy since it allows confirming the diagnosis, analyze the degree of involvement, and determine the anatomical location of the lesion, ruling out differential diagnoses of foot drop, such as peroneal or tibial neuropathy, plexopathy, and radiculopathy. Likewise, the location of the lesion allows us to direct the subsequent imaging study, if necessary. Also, EMG is a useful tool in monitoring, since the presence of signs of re-innervation in the muscles of the sciatic region is a factor of good prognosis. However, it is worth to mention that in the pediatric patient, the procedure should be as limited as possible, since they have less tolerance for it.

While EMG plays a fundamental role in the diagnosis of sciatic neuropathy, the imaging study appears as a tool to determine its etiology and extension, identifying,

for instance, an extrinsic tumor, aneurysm, or abscess. MRI allows the study of sciatic neuropathy, with good soft-tissue resolution and absence of irradiation<sup>22,23</sup>. The muscle MRI that was performed in two cases, showed degenerative changes with a length-dependent pattern (the more distal the muscle, the greater the involvement) in both areas (tibial and peroneal), highlighting again the greater severity in those dependent on the peroneal nerve. MRI has been used both in preclinical research in mice<sup>24</sup> and in humans, as a method to evaluate and even quantify muscle involvement<sup>23</sup> due to denervation in focal neuropathies<sup>22</sup> (including sciatic neuropathy) and in hereditary polyneuropathies<sup>23</sup>. However, its clinical use is still limited. The lumbosacral plexus MRI performed in Patient 6 to evaluate the nerve anatomy, showed a fusiform thickening of the sciatic nerve and the L4 and L5 roots, with increased signal in T2 and a preserved internal fascicular structure, similar to that described in the literature in cases of intraneural perineurioma<sup>25</sup>. The use of MR neurography has been recognized as an effective method to evaluate the anatomy of roots and large nerves, in tumor neuropathies, and inflammatory plexopathies, as well as adjacent structures that could cause compression or infiltration<sup>26,27</sup>. In short, MRI allows us to visualize both the structure of the nerve and adjacent structures, as well as the muscles that are secondarily involved.

With regard to long-term clinical follow-up, motor sequelae and gait impairment were evident in all three cases. A partial recovery is observed in the two patients with neuropathy due to trauma and the patient with neuropathy due to tumor shows no recovery. Patients with neuropathy due to compression and of vascular etiology were not followed-up. Patient 1 did not recover the sensory functionality and partially recovers the motor one, while Patient 2 fully recovers sensory functionality, with residual motor sequelae. Patient 6 does not recover motor function at all, without presenting evident sensory deficits. These findings could be related to the etiology in each one of them (Table 1), with a worse prognosis in the case of tumors than in the case of traumas, being consistent with what has been reported in the literature<sup>1</sup>. In adults, long-term follow-up in patients with iatrogenic sciatic neuropathy has been described as a complication of hip arthroplasty. In mild cases, they presented a full recovery within one year, and in severe cases, 25% recovered completely after five years, 25% had no improvement, and 50% had a partial recovery<sup>24</sup>.

## Conclusion

We presented a group of six girls with sciatic neuropathy, where the main etiologies were due to trauma

or compression, and two of them presented causes not previously described (Hemolytic Uremic Syndrome and Schwannoma/perineurioma). We should suspect sciatic neuropathy in a child who has a foot drop, especially if it is associated with significant neuropathic pain, hip/buttock pathology or trauma, prolonged surgery or altered state of consciousness. We suggest continuing the electrophysiological study with directed EMG that, in this group, allowed the location of the lesion in all cases. Despite the kinesic management, three patients who had long-term follow-up presented significant motor and orthopedic sequelae, therefore, it is essential to prevent this type of lesion in children (accidents, positioning in children with altered state of consciousness). Lumbosacral plexus MRI was key in the diagnosis of one of the patients, while muscle MRI corroborated what was observed in the electrophysiological study and its clinical utility has not yet been defined.

### Ethical Responsibilities

**Human Beings and animals protection:** Disclosure

the authors state that the procedures were followed according to the Declaration of Helsinki and the World Medical Association regarding human experimentation developed for the medical community.

**Data confidentiality:** The authors state that they have followed the protocols of their Center and Local regulations on the publication of patient data.

**Rights to privacy and informed consent:** The authors have obtained the informed consent of the patients and/or subjects referred to in the article. This document is in the possession of the correspondence author.

### Conflicts of Interest

Authors declare no conflict of interest regarding the present study.

### Financial Disclosure

Authors state that no economic support has been associated with the present study.

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