

3. Kuroki S, Saida T, Nukina M, Yoshioka M, Seino J. Three patients with ophthalmoplegia associated with *Campylobacter jejuni*. *Pediatr Neurol*. 2001;25:71–4.
  4. Willison HJ, Yuki N. Peripheral neuropathies and anti-glycolipid antibodies. *Brain*. 2002;125:2591–625.
  5. Kaida K, Kusunoki S. Antibodies to gangliosides and ganglioside complexes in Guillain–Barré syndrome and Fisher syndrome: mini-review. *J Neuroimmunol*. 2010;223:5–12.
  6. Nagashima T, Koga M, Odaka M, Hirata K, Yuki N. Continuous spectrum of pharyngeal–cervical–brachial variant of Guillain–Barré syndrome. *Arch Neurol*. 2007;64:1519–23.
  7. Koga M, Yoshino H, Morimatsu M, Yuki N. Anti-GT1a IgG in Guillain–Barré syndrome. *J Neurol Neurosurg Psychiatry*. 2002;72:767–71.
  8. Kaida K, Kamakura K, Ogawa G, Ueda M, Motoyoshi K, Arita M, et al. S. GD1b-specific antibody induces ataxia in Guillain–Barré syndrome. *Neurology*. 2008;71:196–201.
  9. Odaka M, Yuki N, Hirata K. Anti-GQ1b IgG antibody syndrome: clinical and immunological range. *J Neurol Neurosurg Psychiatry*. 2001;70:50–5.
  10. Santos-García D, Llana M, Macías M, Fuente-Fernández R. Multiple cranial neuropathy associated with herpes simplex virus infection and anti-GM2 immunoglobulin M antibodies. *J Clin Neuromuscul Dis*. 2009;10:199–201.
  11. Susuki K, Koga M, Hirata K, Isogai E, Yuki N. A Guillain–Barré syndrome variant with prominent facial diplegia. *J Neurol*. 2009;256:1899–905.
  12. Ortiz N, Sabaté MM, García N, Santafe MM, Lanuza MA, Tomas M, et al. Effect of anti-GM2 antibodies on rat sciatic nerve: electrophysiological and morphological study. *J Neuroimmunol*. 2009;208:61–9.
  13. Korinthenberg R, Schessl J, Kirschner J, Mönning JS. Intravenously administered immunoglobulin in the treatment of childhood Guillain–Barré Syndrome: a randomized trial. *Pediatrics*. 2005;116:8–14.
- A. Madrid Rodríguez<sup>a</sup>, J. Martínez Antón<sup>a</sup>, M. Núñez Castaín<sup>b</sup>, J.M. Ramos Fernández<sup>a,\*</sup>
- <sup>a</sup> *Sección de Neuropediatría, Servicio de Pediatría, Hospital Regional Universitario Carlos Haya, Hospital Materno-Infantil, Málaga, Spain*
- <sup>b</sup> *Servicio de Neurofisiología, Hospital Regional Universitario Carlos Haya, Hospital Materno-Infantil, Málaga, Spain*
- \*Corresponding author.  
E-mail address: [jmramos@doctor.com](mailto:jmramos@doctor.com) (J.M. Ramos Fernández).

## Levodopa-responsive parkinsonism-dystonia due to a traumatic injury of the substantia nigra<sup>☆</sup>

### Parkinsonismo-distonia unilateral sensible a levodopa por lesión traumática de la sustancia negra

Sir,

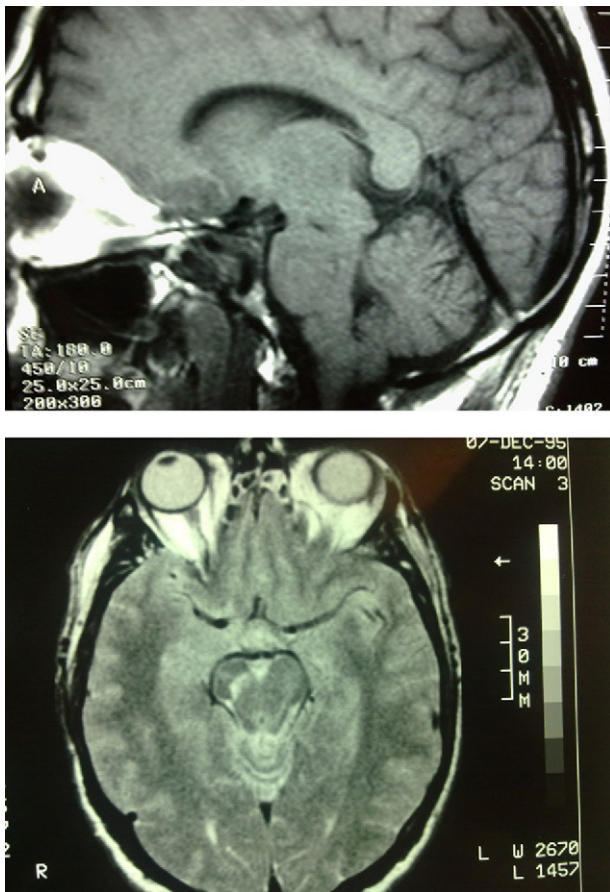
The relationship between traumatic brain injury (TBI) and parkinsonism has been established for a long time.<sup>1,2</sup> However, in exceptionally rare cases there have been reports of parkinsonism secondary to traumatic lesions of the substantia nigra (SN).<sup>3</sup> Case reports of parkinsonism due to SN lesions of vascular origin, either by lacunar stroke<sup>4–7</sup> or by small mesencephalic haemorrhages,<sup>8</sup> are better known, but also exceptional. Bhatt published a series of 3 patients who developed parkinsonism several months after a severe TBI with SN lesions. As characteristic data, there was a delay between the trauma and the onset of parkinsonism. The latter developed quickly and aggressively, with all patients responding to therapy with levodopa.<sup>3</sup> A possible pathophysiological mechanism which was postulated to justify the delay in the onset of parkinsonism with respect to trauma, was iron deposition from the degradation products of the haemorrhagic lesion. This deposition would trigger

the cascade of events typical of dopaminergic degeneration observed in idiopathic Parkinson's disease (IPD), thus explaining the response to dopaminergic therapy in these cases. When the SN is affected, the resulting parkinsonism is strictly unilateral, unless the injury is more extensive and affects other structures. We found very few references in the literature regarding the usefulness of computed tomography or positron emission tomography in this entity, and they mainly referred to cases with a vascular aetiology.<sup>9,10</sup> Recently, a case of parkinsonism secondary to trauma with SN lesion was published in which the transcranial duplex study had not registered hyperechogenicity in the SN, unlike the characteristic pattern in IPD.<sup>11</sup>

We report a case of unilateral parkinsonism-dystonia secondary to traumatic injury of the SN and partially responsive to levodopa. We present the DaTSCAN study, which shows a notable decrease in radioisotope uptake in the striatum ipsilateral to the lesion.

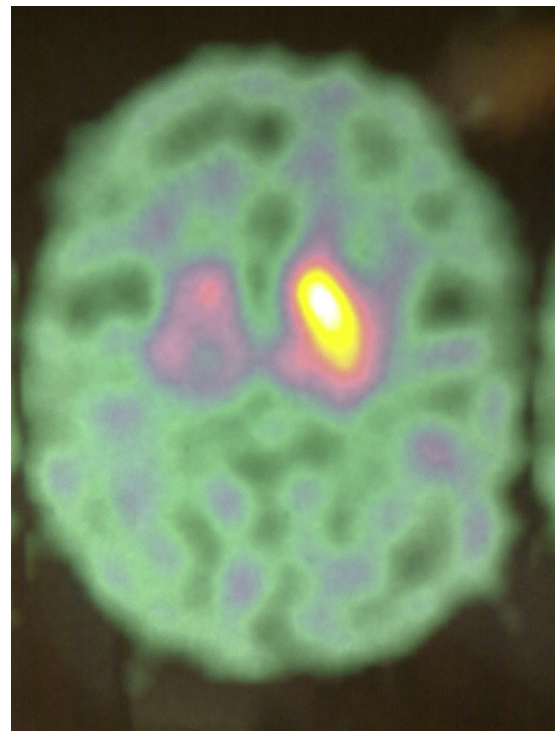
The patient was a 62-year-old male. At the age of 45 he suffered a TBI with loss of consciousness of 30–60 min duration due to a fall from a second storey, with no apparent immediate neurological sequelae. One year later, he started to suffer uncontrolled and involuntary movements of the left limbs, which were more pronounced in the foot. The examination revealed hemidystonia, without any other significant signs. Two months later, in addition to hemidystonia, he suffered akinetic-rigid syndrome characterised by resting tremor, significant cogwheel rigidity and bradykinesia in the affected side of the body. These symptoms had a relatively rapid onset, with severe worsening of parkinsonism within a few weeks. A cranial MRI scan (Fig. 1) conducted at that time showed a right mesencephalic lesion at the level of the SN, with a hyperintense signal on T2-weighted sequences and a hypointense signal

<sup>☆</sup> Please cite this article as: Pérez Errazquin F, Gomez Heredia MJ. Parkinsonismo-distonia unilateral sensible a levodopa por lesión traumática de la sustancia negra. *Neurología*. 2012;27:181–3.



**Figure 1** Cranial MRI scan: sagittal T1 section and axial T2 section. Mesencephalic lesion, hypointense on T1 ("black hole") and hyperintense on T2, at the level of the right *substantia nigra*.

on T1-weighted sequences, and without signal alterations in neighbouring structures (adjacent red nucleus or cerebral peduncle) or at other brain levels. The radiologist reported the lesion as suggestive of residual gliosis in the context of the previous TBI. We conducted an analytical study which included thyroid hormones, Ca, P, Mg, ceruloplasmin, copper, blood smear for evaluation of acanthocytes and serology for syphilis, human immunodeficiency virus and *Borrellia*, with no relevant results. The patient was treated with levodopa/carbidopa at 100/25 mg every 8 h and displayed a partial response, but with a clear improvement over the pre-treatment condition. Since then, his condition has remained stable (approximately 15 years have elapsed since the onset of symptoms), without exacerbation of symptoms or significant worsening or extension to the contralateral side of the body. He has not developed motor complications or dyskinesias associated with levodopa treatment. An attempt to withdraw levodopa was followed by clinical worsening, so it was reintroduced. Hemidystonia persists, but it is mild. At present, the patient continues treatment with levodopa-carbidopa at 300 mg/day and extended-release ropinirole at 16 mg/day. A DaTSCAN was performed recently (Fig. 2), and it showed a notable decrease in uptake in the right striatum and preservation of uptake in the left striatum.



**Figure 2** DaTSCAN image. Marked, unilateral, low uptake of the right striatum.

It is well known that strategic lesions of the striatum or SN can cause symptomatic parkinsonism. The present case behaved like a strictly unilateral parkinsonism, developed months after a TBI. We should note that this parkinsonism occurred after a TBI which was not severe, since there were no immediate and persistent neurological sequelae, as has been previously reported.<sup>3</sup> It started as hemidystonia prior to the development of parkinsonism. Dystonia has been associated with traumatic lesions of the basal ganglia, mostly thalamus and putamen, and also with a delay between trauma and the onset of symptoms.<sup>12</sup> However, there were no lesions in those structures in our case. Recently, a case of hemidystonia due to SN lesion which responded to treatment with levodopa has been published.<sup>13</sup> The cranial MRI scan revealed a lesion with gliotic characteristics at the level of the contralateral SN, with no evidence to justify its presence other than the TBI. Although a vascular lesion cannot be ruled out definitively, the fact is that there were no stroke symptoms at any time, no ischaemic lesions were observed at other levels and the patient presented no cardiovascular risk factors since the event occurred at an early age. In addition, vascular parkinsonism has been associated with an immediate onset after stroke and lack of response to levodopa. These requirements were not fulfilled in our case. Our patient responded to treatment (by this we mean a clear, initial improvement and subsequent stabilisation of symptoms), as described in the few cases reported.<sup>3</sup> No motor fluctuations or dyskinesias associated with prolonged use of levodopa have been detected. We consider unlikely the possibility of IPD precipitated by the TBI, since parkinsonism has remained strictly unilateral after many years of evolution and the DaTSCAN findings are not those of IPD, with complete denervation of one side and preservation

of the other. However, some authors have raised the possibility that TBI could facilitate the development of IPD *a posteriori*.<sup>3</sup> DaTSCAN images verified striatal denervation by Wallerian degeneration following the SN lesion. We emphasise the uniqueness of this case and the novelty of providing a DaTSCAN study.

## References

1. Krauss JK, Jankovic J. Head injury and posttraumatic movement disorders. *Neurosurgery*. 2002;50:927–39.
2. Koller WC, Wong GF, Lang A. Posttraumatic movement disorders: a review. *Mov Disord*. 1989;4:20–36.
3. Bhatt M, Desai J, Mankodi A, Mikhail E, Wadia N. Post-traumatic akinetic-rigid syndrome resembling Parkinson's disease: a report of three patients. *Mov Disord*. 2000;15:313–7.
4. Akyol A, Akyildiz UO, Tataroglu C. Vascular parkinsonism: a case of lacunar infarction localized to mesencephalic substantia nigra. *Parkinson Relat Disord*. 2006;12:459–61.
5. Morgan JC, Sethi KD. Midbrain infarct with parkinsonism. *Neurology*. 2003;60:E10.
6. Ohta K, Obara K. Hemiparkinsonism with a discrete lacunar infarction in the contralateral substantia nigra. *Mov Disord*. 2006;21:124–5.
7. Orimo S, Amino T, Tanaka H, Mitani K, Ishiwata K, Ishii K. A case of hemiparkinsonism following ischemic lesion of the contralateral substantia nigra: a PET study. *Eur Neurol*. 2004;51:175–7.
8. Abe K, Yanagihara T. Hemiparkinsonism following haemorrhage in the contralateral substantia nigra. *Neuroradiology*. 1996;38 Suppl. 1:S67–9.
9. Remy P, De Recondo A, Defer G, Loc'h C, Amarengo P, Plante-Bordeneuve V, et al. Peduncular "rubral" tremor and dopaminergic denervation: a PET study. *Neurology*. 1995;45:472–7.
10. Lorberboym M, Djaldetti R, Melamed E, Sadeh M, Lampl Y. 123I-FP-CIT SPECT imaging of dopamine transporters in patients with cerebrovascular disease and clinical diagnosis of vascular parkinsonism. *J Nucl Med*. 2004;45:1688–93.
11. Kivi A, Trottenberg T, Plotkin M, Niehaus L. Levodopa-responsive posttraumatic parkinsonism is not associated with changes of echogenicity of the substantia nigra. *Mov Disord*. 2005;20:258–60.
12. Kraus JK, Mhadjer M, Braus DF, Wakhloo AK, Nobbe F, Mundinger F. Dystonia following head trauma: a report of nine patients and review of the literature. *Mov Disord*. 1992;1:263–72.
13. Lyoo CH, Lee MS. Levodopa responsive hemidystonia associated with contralateral nigral lesion. *J Neurol*. 2010;257:856–7.

F. Pérez Errazquin\*, M.J. Gomez Heredia

*Unidad de trastornos del movimiento, Servicio de Neurología, Hospital Universitario Virgen de la Victoria, Málaga, Spain*

\*Corresponding author.

*E-mail address:* pacoerrazquin@hotmail.com

(F. Pérez Errazquin).

## The transfer of neuropaediatrics to adult medicine<sup>☆</sup>

### La transferencia de neuropediatría a medicina de adultos

Sir,

Neuropaediatricians manage patients from the neonatal period through adolescence and youth. Whenever their patients are not discharged, sooner or later they must be transferred to adult physicians: family physicians, neurologists, neurosurgeons or other specialists.

This transfer is necessary because neuropaediatricians are experts in children and adolescents but not in adults. Moreover, it is inappropriate for adults to be treated in paediatric wards.

However, this transfer can be traumatic because, in some cases, a relationship has existed for many years. This includes both the way of working and the affection established between neuropaediatricians and patients and their families, who must adapt to new environments and new

professionals. Furthermore, neurologists are not always prepared to deal with some neuropaediatric diseases.

The issue of transfers was raised for the first time in the United States, at a convention which took place in 1984, as a result of the significant increase in survival in children with chronic and disabling conditions during the 1970s and 1980s. Subsequently, there have been 2 other conventions in 1989 and 2001.<sup>1</sup>

A search in PubMed, dated February 13th, 2011, for reviews containing the terms "pediatric to adult transition", returned 130 articles (of which only 8 were from the 20th century) related to cystic fibrosis, cancer, transplants, congenital heart disease, diabetes mellitus, inflammatory bowel disease, chronic kidney disease, growth hormone deficiency and neurological and neurosurgical pathology.

In total, 12 articles made reference to neurological problems concerning unspecified neurological diseases,<sup>2</sup> miopathies,<sup>3</sup> hydrocephalus,<sup>4</sup> CNS tumours,<sup>5</sup> attention deficit hyperactivity disorder (ADHD),<sup>6</sup> neurosurgical pathology,<sup>7–9</sup> brain trauma,<sup>10</sup> phacomatosis and genetically determined tumours,<sup>11</sup> spina bifida<sup>12,13</sup> and cerebral palsy.<sup>13</sup> In addition, 1 review made reference to inborn errors of metabolism,<sup>14</sup> many of which have a neurological effect. Until recently, their diagnosis and follow-up corresponded to paediatricians, but, at present, adult services (especially neurology and internal medicine) must also adapt to them.<sup>15</sup>

All the articles agree that very little progress has been made and highlight a number of barriers for a successful transition: lack of coordination between paediatric and

<sup>☆</sup> Please cite this article as: López Pisón J, et al. La transferencia de neuropediatría a medicina de adultos. *Neurología*. 2012;27:183–5.