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Case report

Long-term follow-up of an adolescent who had bilateral striatal necrosis secondary to *Mycoplasma pneumoniae* infection

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Abstract

We describe the long-term follow-up of a patient with bilateral striatal necrosis associated with *Mycoplasma pneumoniae* infection occurred in adolescence. In the literature there are no longitudinal studies of such a rare condition. Our patient, 4 years after the onset of an acute and reversible akinetic-rigid syndrome, showed a severe obsessive-compulsive disorder, cognitive decline, and a neuropsychological profile characterized by signs of deficient executive functioning. The clinical picture that emerged in our patient is suggestive of a frontosubcortical dementia which might be considered a major long-term sequela of the bilateral selective striatal necrosis and consequent dysfunction of frontostriatal connections.
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Keywords: Bilateral striatal necrosis; *Mycoplasma pneumoniae*; Frontosubcortical dementia; Obsessive-compulsive disorder

1. Introduction

Obsessive-compulsive disorder (OCD) has frequently been linked with neurological diseases primarily affecting the basal ganglia. Both anatomical and neuroradiological studies suggest that OCD could be related to a dysfunction of frontostriatal connections [1].

In a previous paper we discussed the case of an adolescent patient who presented a reversible akinetic-rigid syndrome associated with selective striatal necrosis related to a *Mycoplasma pneumoniae* infection focusing particularly on clinical and MRI aspects of the acute phase of the disease [2]. As in the literature there are no longitudinal studies of such rare cases we now report the long-term follow-up of the same patient who presented cognitive and obsessive-compulsive behavioural changes 4 years after the onset of the first symptoms.

2. Case report

In March 1997, a previously healthy, 17-year-old boy developed a febrile illness. Over the next 4 days he showed a progressive akinetic-rigid syndrome. The MRI revealed cytotoxic oedema restricted specifically to both the striata; no contrast enhancement was observed after gadolinium-DTPA administration (Fig. 1). Chest X-ray was consistent with atypical interstitial pneumonia. The serum and CSF antimycoplasma antibody titers were high (IgG and IgM). Treatment with erythromycin, rifampicin and L-dopa combined with carbidopa was started. One month after the onset of symptoms, we observed progressive improvement of the patient's neurological condition, which persisted after discontinuation of the L-dopa treatment. After 4 months, neurological examination revealed only choreiform movements of the upper extremities and mild dystonic postures of the feet. Follow-up MRI studies were performed 20 days, and 2, 3, 5, and 9 months after the onset of symptoms and documented the onset of striatal atrophy and compensatory enlargement of the frontal horns of the lateral ventricles. The nuclei showed an abnormally increased signal intensity

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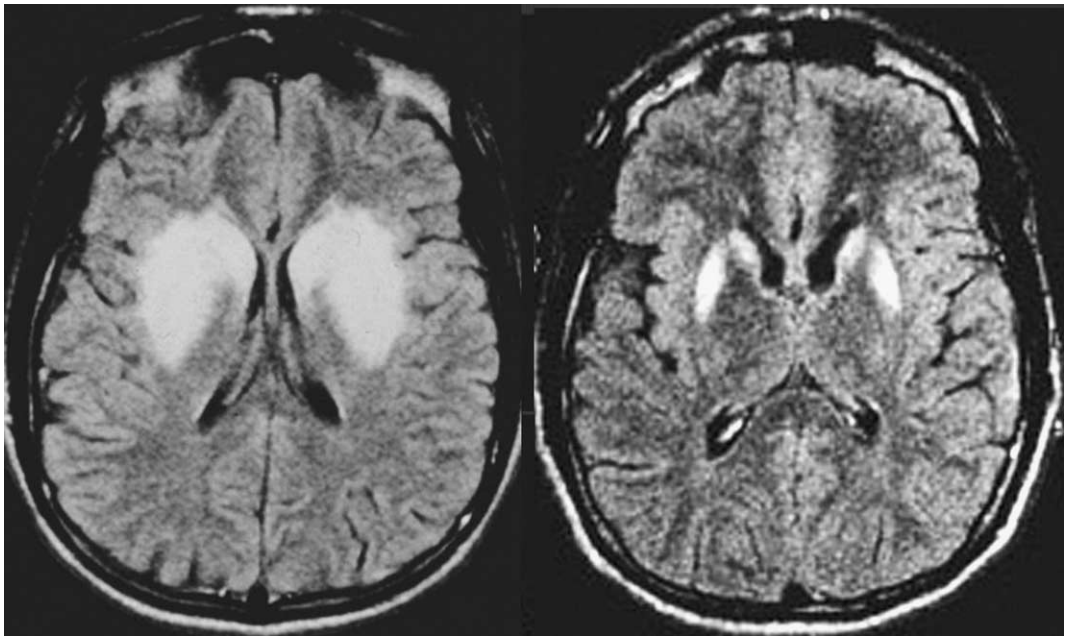

Fig. 1. Axial FLAIR images (TR: 11000 TE: 140) shown on the left, during the acute phase, marked swelling and abnormally increased signal intensity of the putamen and caudate nuclei bilaterally. On the right, the MRI performed 9 months later, demonstrates bilateral striatal atrophy and persisting high signal intensity.

in T2-weighted images, more marked in the heads of the caudate nuclei and in the anterior portion of the putamen.

In our earlier report [2], we concluded that the finding of severe striatal alteration on MRI did not seem to correlate with our patient's favourable clinical course. He returned for a follow-up visit in March 2001 (4 years after the onset of the first symptoms) as, for several months, he had been experiencing a growing restlessness and marked hyperkinesia: a compulsive need to walk for a number of hours a day. These symptoms were accompanied by obsessive ideas (relating to China, the Taliban, and Maradona), and a tendency to talk repeatedly about these subjects with anyone he met. The obsessions and compulsions reported by the patient fulfilled the DSM-IV criteria for a diagnosis of OCD, very severe according to the Yale-Brown Obsessive Compulsive Scale (Y-BOCS) (Table 1). The neurological examination revealed a marked akathisia and slight extrapyramidal signs at buccolingual and lower limb level (action dystonia). Routine blood tests and EEG gave normal findings. Brain MRI after the 9 months showed atrophy and gliosis of the striata (Fig. 1).

The patient underwent an in-depth neuropsychological evaluation to assess attention, intelligence, language, memory and executive functions. The tests administered are listed in Table 1. The results were normalized to z scores to allow direct comparison of tests and are summarized in Fig. 2 (by definition the z score of the normal population is zero and has an SD of 1). At the first observation (4 years earlier), it had not been possible, due to the severity of the patient's clinical conditions, to administer any neuropsychological tests, whereas shortly afterwards (only 8 months after the onset of the first symptoms), we were able to perform a cognitive evaluation using the WAIS scale: this gave a full-scale IQ of 79 (z score -1.4), a verbal IQ of 85 (z score -1) and a performance IQ of 79 (z score -1.4), as summarized in Fig. 2. The WAIS scale was once again administered at the most recent follow up and revealed a cognitive decline: this time, it gave a full-scale IQ of 70 (z score -2), a verbal IQ of 73 (z score -1.8) and a performance IQ of 70 (z score -2). While language and attention were unaffected, deficits were observed in short-term verbal memory and verbal fluency.

The results of all the tests administered, summarized in Fig. 2, delineate a neuropsychological profile suggestive of frontosubcortical dementia.

3. Discussion

In our patient, 4 years after a bilateral striatal necrosis associated with *M. pneumoniae* infection, we found a picture of severe OCD, cognitive decline and a neuropsychological profile characterized by signs of deficient executive functioning. In the literature there are no longitudinal studies of bilateral striatal necrosis associated with *M. pneumoniae* infection in the pediatric age group. The already published reports of such rare cases (Green and Riley [3] reported one case and reviewed six previously reported cases including our one [2]) described the acute symptomatology of the patients and not the long-term sequelae of the disorder. Our patient is affected by a possibly immune-mediated and therefore selective striatal lesion that makes him particularly interesting from the perspective of speculation over the possible functions of

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