Prism adaptation improves spatial dysgraphia following right brain damage

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Received 7 September 2005; received in revised form 27 March 2006; accepted 2 April 2006
Available online 18 May 2006

Abstract

Visuo-manual adaptation to prisms produces a long-lasting improvement of visuo-spatial neglect. Improvement is also observed in tasks that do not involve visuo-manual component and that can all be considered to rely on a rightward (ipsilesional) orienting bias. Here, we report positive effects of prism adaptation on spatial dysgraphia, in a neglect patient following right brain damage. A long-lasting improvement concerned the top-down preference reflecting the ipsilesional bias but also the sloping lines and the broken lines reflecting visuo-constructive disorders in handwriting. Moreover, a transient improvement was also evidenced for the graphic errors. These results reinforce the idea that the process of prism adaptation may activate brain functions related to multisensory integration and higher spatial representations and show a generalization at a functional level. Prism adaptation therefore appears as useful tool in the theoretical attempt to identify the underlying 'core' mechanisms of the neglect syndrome.

Keywords: Neglect; Spatial dysgraphia; Prism adaptation; Visuo-constructive disorder; Right hemisphere; Rehabilitation

1. Introduction

Hemispatial neglect may be considered as a failure to generate or maintain a normal representation of the left side of space (Bisiach, Luzzatti, & Perani, 1979) that expresses as a failure to report, respond to, or orient toward novel and/or meaningful stimuli of this space (Heilman, Watson, & Valenstein, 1985). Far from being a unitary condition, hemispatial neglect is a protean disorder whose symptoms can selectively affect different sensory modalities, cognitive processes, spatial domains and coordinates systems (Halligan, Fink, Marshall, & Vallar, 2003). Interestingly many of these different manifestations may be alleviated following visuo-motor adaptation to right-deviating prisms (Rossetti et al., 1998); the improvement concerns not only the visuo-spatial domain and egocentric reference frame (Frassinetti, Angeli, Meneghelli, Avanzi, & Ladavas, 2002; Rossetti et al., 1998) but also the representational space (Rode, Rossetti, & Boisson, 2001; Rossetti et al., 2004) and other sensory modalities like audition or touch (Dijkerman et al., 2003; Maravita et al., 2003; McIntosh, Rossetti, & Milner, 2002). Therefore, these effects cannot be explained by a simple sensorimotor post-effect and rather suggest a cognitive post-effect induced by prism adaptation.

Functionally, prism adaptation has been shown to improve disabling deficits of everyday life activities like postural balance (Tilikete et al., 2001), reading (Farnè, Rossetti, Toniolo, & Ladavas, 2002) and wheelchair driving (Courtois-Jacquin et al., 2006). Another disabling deficit observed in right-brain damaged patients with hemineglect concerns the handwriting (Rode, Perenin, & Boisson, 1996). In their more severe expression, handwriting difficulties have been qualified as spatial dysgraphia (Ardisia & Rosselli, 1993; Cubelli, Guiducci, & Consolmagno, 2000; Hecaen, Angelegues, & Douzenis, 1963; Hecaen & Marcie, 1974; Lebrun, 1985). Spatial dysgraphia has been defined as “a disturbance of graphic expression due to an impairment of visuo-spatial perception resulting from a lesion in the nonlanguage-dominant hemisphere” (Hecaen & Albert, 1978, p. 66). According to these authors, four main features...
define spatial dysgraphia—(i) “right-page’ preference: writing is crowded onto the right side of the page leaving an excessively wide “margin” on the left side; (ii) sloping lines (inclination): patients fail to write horizontally and produce oblique or wavy lines; (iii) broken lines: patients leave abnormally large space between words, thus leading to the fragmentation of the line into small segments; (iv) graphic errors: production of an incorrect number of strokes for a given letter or of letters for a given word. The present article reports the first investigation of the effect of prism adaptation on spatial dysgraphia.

The heterogeneous manifestations of left hemispatial neglect may be classified in two categories: those that can be explained by a right-oriented behavioural disorder and the others, dynamic and visuo-constructive disorders in nature, that cannot (Pisella & Mattingley, 2004). The question of the effect of prism adaptation on the second category of disorders is still open. Clinically, these two categories of disorder are frequently associated, in particular in spatial dysgraphia. Indeed, the “right-page” preference reflects the right-oriented bias while the inclination of lines, the gaps and the graphic errors may be considered different in nature, perhaps as visuo-constructive errors. Prism adaptation clearly induces a reduction of the right orienting bias that has been measured in straight-ahead pointing (e.g. Pisella, Rode, Farnè, & Boisson, 2003, Fig. 6, p. 284; Rossetti et al., 1998; Tilikete et al., 2001), visuo-graphic tasks like cancelation or copy (Rossetti et al., 1998) and mental imagery (Rode et al., 2001; Rossetti et al., 2004). In addition, the symmetry (object-based neglect) and the spatial organisation of drawing has already been reported to improve following prism adaptation in individuals e.g. in the Gainotti test (Rode, Pisella, Rossetti, Farnè, & Boisson, 2003, Fig. 6, p. 284; Rossetti et al., 1998; Fig. 6 p. 284), suggesting a possible effect of prism adaptation on visuo-constructive disorders. An improvement of the four features of spatial dysgraphia described above could thus be predicted. The aim of this study was to test the potential effect of prism adaptation on each of the four parameters of spatial dysgraphia.

2. Methods

2.1. Case description

A 69-year-old, right-handed man (R.C.) was admitted to hospital in October 2001 for acute left hemiplegia secondary to a vascular infarct in the territory of the right middle cerebral artery (Fig. 1). A CT-scan performed one month post-onset showed a temporal-patalial infarct including internal capsule, putamen and corona radiata.

The patient initially had a severe left upper and lower limbs paresis with a tactile sensory deficit and an astereognosis. Speech was mildly dysarthric due to a left central facial paresis. On Goldmann’s perimetry there was a left superior quadrantanopia. Neuropsychological examination at one month post-onset revealed a head and gaze deviation toward the right side and a marked left-sided neglect. The patient omitted most of the left-sided items and exhibited visuo-constructive errors when asked to draw from memory or copy a daisy, a house or a bicycle. On a line-crossing task (Albert, 1973), the patient omitted all the segments of the leftmost column (25/40). When required to bisect horizontal lines, he significantly transacted to the right of the centre. When asked to touch his left hand with the right, he reached the target with hesitation and search, (i.e. he got a score of 1 on the four-point scale of Bisiach, Petros, Vallar, & Berti, 1986, for personal neglect). He omitted to shave the left half of his face and sometimes also forgot to slip on the left side of his clothes. The patient had no aphasic deficit, was well oriented in time and exhibited no mental deterioration. The patient also displayed a marked spatial dysgraphia. When asked to write on an A4 paper placed in front of him under dictation or by copy, the leftmost part of the sheet was neglected. Others disturbances were noted: an inclination of lines with an attraction of written text to the right and superior cost of the sheet, some lines broken with an inability of the patient to follow a horizontal line. Lastly, analysis of writing revealed several errors as omissions or repetitions. By contrast, the patient exhibited a minor spatial dyslexia with only few omissions in the left space of the page during a task of single word or text reading.

At the time of examination, the patient did not show personal neglect and anosogasia had disappeared, the patient was quite alert, well oriented in time and cooperative. The experimental procedure was applied one month post-onset.

2.2. Prism adaptation

2.2.1. Apparatus

The apparatus used in this experiment is similar to that employed by Redding and Wallace (1996). Specifically, this structure consisted of a two-layer rectangular black wooden box-like frame (30 cm high, 80 cm wide and 80 cm deep) placed on a table and opened on the side facing the subject. Throughout the experiment, the patient was seated in front of the structure and placed his arms
within the structure. Their head was kept aligned with the body sagittal axis using a chin-rest situated on the top of the box. A piece of foam placed on the lower layer of the box near the sternum of the patient indicated the hand starting position. Sight of the starting hand position and of the first approximately 15 cm of the pointing surface was not permitted by the structure. The last part of their pointing trajectory remained visible throughout the adaptation procedure. Throughout the experiment, the patient was required to point on the lower surface of the box and return to the starting position after each trial. The lower horizontal surface of the box was covered by electroconductive paper that allowed the movement endpoints to be recorded via a metallic thimble attached to the tip of a subject’s index finger. This apparatus produced measurements with an accuracy of 0.1°.

2.2.2. Adaptation procedure

The patient was exposed to a shift of the visual field to the right produced by prismatic lenses. The goggles were fitted with wide-field, wedge lenses creating an optical shift of 10°. The exposure period consisted in 50 pointing responses to visual targets presented 10° to the right or to the left of the objective body midline. During the prism exposure, the patient was asked to point at a fast but comfortable speed; they could see the target, the second half of their pointing trajectory and their terminal error. Their head was kept aligned with the body’s sagittal axis by a chin-rest and controlled by an investigator. The total duration of this exposure was about 3 min (see details in Rossetti et al., 1998).

2.2.3. Straight-ahead pointing test

The patient was seated blindfolded in front of the horizontal box. He was required to point straight-ahead while his head was kept aligned with the body’s sagittal axis. Ten pointing trials were performed in the pre-test (without prisms) and in the post-test (immediately upon the prism removal), in order to check that prism adaptation had been efficient. Mean deviations observed before and immediately after prism adaptation were compared by t-test.

2.3. Neuropsychological assessment

2.3.1. Procedure

The performance of the patient on line bisection, free drawing and writing was repeatedly evaluated over six sessions within a time period of four consecutive days. These three tasks were administered one day before (~24 h), just before (~2 h), just after the short prism adaptation session (~0 h), 2 h after, then again 24 h and 48 h after.

2.3.2. Assessment of neglect

Unilateral spatial neglect was assessed free drawing and line bisection. The patient was asked to draw a daisy and a clock from memory and to mark the midline of 20 horizontal lines presented in the right, centre and left part of the same testing sheet (Schenkenberg, Bradford, & Ajai, 1980). This sensitive test allowed us to compute a score reflecting the bias in line bisection as proposed by Schenkenberg et al. (1980). It expresses the mean percentage of horizontal deviation on the line bisections, towards the right (signed positive) or towards the left (signed negative).

A one-way factorial ANOVA was performed with the three periods of testing (Pre-adaptation, Immediately Post-adaptation and Late after adaptation) as factors and the writing task (daisy and o’clock) as the dependent variable. The area of the page not used to write was measured in mm² and then expressed in percentage of the surface of the sheet.

2.3.3. Assessment of handwriting

The writing performance was assessed under two different conditions: writing from dictation or copy. In the writing-from-dictation task, the patient was asked to write thirty eight sentences (68 words, ranging from 2 to 9 letters) from a poem entitled ‘Le chemin creux’ by Verlaine on a visiting card (7.9 cm × 11.2 cm). The two texts were presented to the patient in cursive style from a poem entitled ‘Le chemin creux’ by Verlaine on a visiting card (7.9 cm × 11.2 cm). The two texts were presented to the patient in cursive style from a poem entitled ‘Le chemin creux’ by Verlaine on a visiting card (7.9 cm × 11.2 cm). The two texts were presented to the patient in cursive style from a poem entitled ‘Le chemin creux’ by Verlaine on an A4 paper sheet and secondly four sentences (28 words, ranging from 2 to 10 letters) from a ‘poème surmerveillé’ by Verlaine on a visiting card (7.9 cm × 11.2 cm). The two texts were read aloud by the examiner.

In the copying task, four printed sentences (33 words, ranging from 2 to 15 letters) were presented to the patient in cursive style from a poem entitled ‘Le chemin creux’ by Verlaine on an A4 paper sheet. In this task, the model was placed above the paper sheet and in the midsaggital plane. The four following parameters of spatial dysgraphia were measured (see Fig. 2):

- Right-page preference: The surface of the page neglected by the patient (the area of the page not used to write) was measured in mm² and then expressed in percentage of the surface of the sheet.
- Broken lines: The number of abnormal spaces inserted between words was counted. These spaces often resulted in “steps” in the written line.
- Sloping lines: The angle of inclination of the lines of writing (to the top or bottom of the page) was measured comparatively to the horizontal (in degrees). A positive value indicates a top inclination and a negative value a down inclination.
- Graphic errors: The number of omissions, of duplications of strokes, letters or words and of abnormal spaces between letters was added.

Given the differential variation ranges between parameters and between format (A4/name-card), statistics have been realised on data expressed in per-
3. Results

3.1. Straight-ahead demonstration

In the pre-test, the straight-ahead pointing was initially shifted from the midline toward the ipsilesional side (mean: 16.2°; standard error of the mean (S.E.M.): 2.6). In the post-test, R.C. demonstrated a reduction of straight-ahead shift (mean: 1.4°; S.E.M.: 3.6). Comparison by t-test of mean deviation observed before and following the adaptation revealed a highly significant difference (t(1,9) = 7.9; p < 0.0001) indicating that the patient showed adaptive after-effects to a lateral shift of the visual field to the right.

3.2. Line bisection test

For the line bisection test, in the two pre-tests, the deviation scores of R.C. were, respectively, 37.5% and 31.8%. In the post-tests the deviation scores were, respectively, 25.7% in the immediate post-test, 29.4% at 2h, 24.6% at 24h and 20.2% at 48h. Performance of R.C. was significantly improved after adaptation (F(1,137) = 5.15; p < 0.05 between pre and post-values). However, note that this same test has been performed again one week later on occasion of routine clinical assessment with a performance similar to the pre-test (deviation score of 41.1%, F(1,137) = 1.23; p < 0.3).

3.3. Free drawing from memory

For the free drawing of object, in the pre-test, R.C. neglected the left half of a daisy or a clock drawn from memory. In the post-

![Graphs of different parameters of spatial dysgraphia](image_url)
tests, the patient drew an entire daisy with symmetrical number of petals and a symmetrical clock (see Fig. 2).

3.4. Handwriting

For each of the four parameters measured to evaluate the writing deficits, the factorial ANOVA did show a significant main effect of the period of testing (Pre, Immediately Post and Late after prismatic adaptation) on the writing performance (all $F(2,9) > 4$, $p < 0.05$). By contrast, no main effect of the conditions of testing (Copy on A4, Dictation on A4 and Dictation on a name-card) was revealed by the statistical analysis, for none of the four parameters (all $F(2,9) < 1.7$, $p > 0.05$). No significant interaction was revealed between the two factors. Evolution of each parameter of writing deficit is thus illustrated in Fig. 3 as a mean measure of the three conditions of testing with respect of testing time (in hours). Periods of testing are mentioned on the graphs.

For the right-page preference: planned comparisons show a significant short-term (Pre- versus Post-Immediate: $F(1,9) = 8.16$, $p < 0.05$) and long-term effect (Pre versus Late: $F(1,9) = 13.62$, $p < 0.05$) of prism adaptation on the neglected surface area.

For the broken lines: planned comparisons show a significant short-term (Pre- versus Post-Immediate: $F(1,9) = 26.08$, $p < 0.05$) and long-term effect (Pre versus Late: $F(1,9) = 24.30$, $p < 0.05$) of prism adaptation on the neglected surface area.

For the right-page preference: planned comparisons show a marginally significant short-term (Pre- versus Post-Immediate: $F(1,9) = 4.98$, $p = 0.05$) effect but a long-term effect (Pre versus Late: $F(1,9) = 12.63$, $p = 0.05$) of prism adaptation on the neglected surface area.

For the sloping lines: planned comparisons show a marginally significant short-term (Pre- versus Post-Immediate: $F(1,9) = 1.7$, $p > 0.05$) and long-term effect (Pre versus Late: $F(1,9) = 24.30$, $p < 0.05$) of prism adaptation on the neglected surface area.

Lastly for the graphic errors: planned comparisons show a significant short-term (Pre- versus Post-Immediate: $F(1,9) = 8.05$, $p < 0.05$) effect but no long-term effect (Pre versus Late: $F(1,9) = 0.4$, $p > 0.05$) of prism adaptation on the number of graphic errors.

4. Discussion

The aim of this study was to assess the effects of prism adaptation on spatial dysgraphia, a disabling deficit frequently associated to neglect following right brain damage. Moreover, this constituted a mean to test whether prism adaptation could reduce the visuo-constructive disorders related to the right hemisphere function, as it had been shown already for the leftward orienting behavioural biases of the neglect syndrome.

First, the results did confirm the positive effect of prism adaptation on the oriented behavioural disorder, as previously reported (e.g. Pisella et al., 2002; Rode et al., 2003; Rossetti et al., 1998). In the present paper, it is clearly demonstrated by the reduction of the ipsilesional shift in the straight-ahead pointing in darkness and in the line bisection task and by the decrease of the neglected surface. The reduction of the neglected area also persisted in the late test (48 h), confirming the potential long-lasting improvement of this category of disorder (Farn`e et al., 2002; McIntosh et al., 2002; Pisella et al., 2002). The results secondly revealed the reduction of the lines inclination and of the number of broken lines, maintained 48 h later, as well as the transient decrease of graphic errors after prism adaptation. This rehabilitation technique is therefore able to induce a significant improvement of handwriting in neglect patients. The ability to place and orient correctly the words in the page and relatively to each others was durably improved; the decrease of the graphic errors observed within words was only observed immediately after the prism removal. The improvement of these features of spatial dysgraphia is hardly explained by the leftward-oriented sensori-motor after-effect of prism adaptation. The positive effect of prism adaptation on visuo-constructive disorders suggests that prism adaptation does not act specifically on the ipsilesional bias characteristic of hemineglect but rehabilitates more generally the visuo-spatial functions attributed to the right cortical hemisphere. Prism adaptation may enlarge or shift the part of space represented in the spared cortical hemisphere (see Redding & Wallace, 2006). This different distribution of the representation of space between hemispheres could explain the improvement of both the right-oriented behavioural disorders and the visuo-constructive disorders.

The short-term effect of prism adaptation on the graphic errors can lead to rather distinguish this latter parameter from the other parameters which were still improved after 48 h. Accordingly, the graphic errors of spatial dysgraphia have been often claimed in the literature to result from a disorder distinct from the oriented behavioural bias and the visuo-constructive disorder. The definition of graphic error has however not always been consistent in these papers. In the present paper, graphic errors did include iterations (duplications of strokes, letters or words) as well as omissions and abnormal spaces between letters and words (Hecaen & Albert, 1978).

Ardila and Rosselli (1993) have comparatively studied handwriting deficits in retro-rolandic and pre-rolandic right hemisphere damaged patients. In a group of 21 patients, about half of pre-rolandic and three-quarters of retro-rolandic presented with writing errors and spatial dysgraphia of variable degrees. For further investigations of handwriting performance, right hemisphere damaged patients were selected on the presence of spatial dysgraphia but exhibited associated disorders including left hemiparesis, visual field defects, spatial hemineglect, constructional apraxia, spatial alexia or spatial acalculia. Iterations have been considered the most distinctive type of writing error in right hemisphere damage patients (Hecaen & Marcie, 1974) and neglect has been considered partially responsible for iterations of strokes and letters: once the stroke and letter is written, the patient neglects it, since it moves to the left visual field (Simmenskaya, 1974). However, Ardila and Rosselli (1993) established that neglect did not correlate with iterations and that iterations, when produced during writing, are also observed when drawing figures and the information remains in the same absolute position in the visual field. On the other hand, feature additions did significantly correlate with left hemiparesis and hence pre-rolandic damage. Altogether, the correlation study suggested that writing deficits in right hemisphere damaged patients can be associated to and depending on four components: (1) left hemi-neglect, reflected in increased and unsteady mar-
gins; (2) constructional deficits, reflected in changes in the type of handwriting and misgroupings of elements; (3) general spatial deficits, reflected in the inability to use correctly the spaces between words, difficulty in keeping the written line in a horizontal position, disrespect to spaces and spatial disorganization of the written material; (4) a tendency to perseverate, reflected in iterative errors. Although all types of errors were produced in both groups of patients and were more numerous in patients with retro-rolandic damage (not only in writing words and sentences, but also in writing letters and syllables), the authors put forward that iterative errors (feature additions and letter additions) represented the most evident type of writing defect in the right pre-rolandic patients, whereas misgrouping of elements and letter omissions represented the two most evident types of writing errors in right retro-rolandic patients.

Lebrun and Rubio (1972) have argued that errors as duplications and omissions of strokes and letters correspond to “afferent-type” errors because these errors are similar to those made by normal subjects when requested to write with their eyes closed or with delayed visual feedback. The patients producing these afferent-type errors may add a fourth downstroke to the letter ‘m’ or omit the second downstroke of the letter ‘n’ (Lebrun, 1976). These errors occur mainly in writing words with double letters or in writing letters with repeated strokes (Hecaen & Marcic, 1974; Margolin, 1984). However, unlike normals, dysgraphic patients are reported not to be more defective when vision is prevented (Lebrun, 1976), the term “afferent dysgraphia” was thus used to refer to the inability to use afferent information in monitoring writing movements. Some pure forms of afferent dysgraphia with no evidence of left visuo-spatial neglect following a focal damage of the right hemisphere have been reported (Crossile & Hibert, 1998; Cubelli & Lupi, 1999). Moreover, Cubelli et al. (2000) have studied the writing performances of a series of nine right hemisphere damaged patients with a written production disorder and shown that afferent-type errors may be clearly dissociated from neglect-related writing deficits and spatial errors. For these authors afferent dysgraphia may be even considered as an autonomous syndrome, distinct from the spatial dysgraphia picture. The neglect-related and spatial writing errors should reflect an oriented behavioural disorder and/or a visuo-constructive disorder although the afferent errors should reflect rather a disorder in monitoring the actual handwriting movements in order to compare what is written with the abstract representation of letters (Cubelli et al., 2000).

More recently, Pisella and Mattingley (2004) have listed a number of manifestations that cannot be explained by current theories of neglect, and especially by the dominant attentional theory that solely postulates an orienting bias toward the side of lesion (ipsilesional bias), a local bias and impaired sustained attention. They have postulated that these manifestations, mostly visuo-constructive in nature, can be accounted for by an additional underlying disorder of spatial remapping due to parietal dysfunction. In primary visual areas, retinotopic maps are renewed and thus overwritten at each new ocular fixation. Remapping processes operating in higher-level oculocentric maps of the parietal cortex ensure visual integration of these successive retinal images over time and space. More specifically, the authors explained that in right brain damaged patients a remapping impairment would result in an overwriting from visual conscious representation of the visual space opposite to the visual orientation, which is usually rightward due to the ipsilesional bias. The overwritten left visual space is defined with respect to the previous eye position, i.e. before the saccade occurred. The visual integration of successive retinal images appears particularly important during handwriting, which involves multiple eye movements and a constant monitoring and on-line integration of which part of the sentences, words and letters has been already written and which part remains to be written, in what way (inclination) and where (gaps). With an impairment of remapping mechanisms, the successive rightward saccades involved during handwriting would lead to progressive overwritten of the left space, corresponding to the part of the sentence that has already been produced. This would therefore affect the on-line visual monitoring of handwriting and lead to errors like duplications of words, letters or strokes, incorrect inclinations, gaps between words and broken lines, as if the handwriting was done with eyes closed. Consistent with this hypothesis, there would be no reason to distinguish errors due to this “visual” monitoring deficit occurring within words (graphic errors) or between words (inclinations and gaps/broken lines). However, in case of frontal dysfunction (that the infant, which included the internal capsule, putamen and corona radiate, suggests may be present in the patient reported in this paper), handwriting errors of a perseverative nature can be exhibited (Ardila & Rosselli, 1993; Rusconi, Maravita, Bottini, & Valler, 2002), especially within words, probably due to impaired “motor” monitoring between the actual handwriting movements produced and the abstract representation of letters (Cubelli et al., 2000). The present results would suggest that the former, spatial visuo-constructive errors, can be durably improved by prism adaptation, whereas this is not true of the latter, perseverative graphic errors. Our patient has both a parietal and a frontal lesion. The absence of long-term improvement of graphic errors suggests that these can be considered as a frontal perseverative dysfunction in our patient, whereas the other dysfunctions can be considered as visuo-constructive in nature and caused by the parietal dysfunction. Although spatial dysgraphia that possesses all of the four main features of right-page preference, line inclination, broken lines and graphic errors, as indicated by Hecaen et al. (1963), is a rare clinical condition, it is never clear to what extent one can generalize from single case studies. For this reason, it is important to note that further studies of similar cases will be necessary to confirm whether the interpretation offered here is correct.

Since the spatial deficits consecutive to right brain damage are still weakly defined and have almost all been shown to be possibly dissociated, the symptoms of spatial dysgraphia and hemispatial neglect have been classified in multiple manners and divided into more or less numerous components. These attempts of classifications are interesting because of their theoretical and/or experimental work in order to identify the underlying ‘core’ mechanisms of these multiple manifestations, if any. Although discrepancy in the duration of the amelioration of given tests has already been observed within and between individual patients in the literature (e.g. Pisella et al., 2002; review...
in Rode et al., 2003), the potential modification of the postulated components by prismatic adaptation appears as an useful additional test of the various classifications.

Acknowledgments

The authors wish to thank J.L. Borach for his work on the figures. This work was supported by ACI Neurosciences (Plasticité), Leverhulme Trust and INSERM PROGRES.

Parts of this study were presented to the European Congress of Neuropsychology, 18-20 avril 2004, Modena, Italy.

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