

PRISM ADAPTATION IMPROVES CHRONIC VISUAL AND HAPTIC NEGLECT: A SINGLE CASE STUDY

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ABSTRACT

Visuomotor adaptation to rightward displacing optical prisms is known to induce temporary improvements in the symptoms of left visual neglect. We report a 74 year-old woman with severe and chronic neglect of nine months duration, who underwent three weekly sessions of prism adaptation. Substantial improvements were obtained on tests of visual neglect (cancellation, copying and bisection). Improvement was also observed on a spatial judgement task, with no explicit visual component, in which CS was required to locate the centre of a haptically explored circle. These observations confirm that brief periods of prism exposure can benefit even chronic neglect disorders. Moreover, the improvement observed on the haptic task supports the belief that this procedure can influence higher levels of spatial representation.

Key words: haptic, neglect, prism adaptation, rehabilitation

INTRODUCTION

Hemispatial neglect is a common syndrome in which a patient fails to report or respond to stimulation from the side of space opposite a brain lesion (usually of the right hemisphere), where these symptoms are not due to primary sensory or motor deficits (Heilman, 1979). The syndrome has received intensive study over the last two decades for a number of reasons. First, it provides a fascinating window on the organisation of spatial representation in the brain. The fact that neglect is widely heterogeneous, manifesting many dissociable subtypes, implies some degree of functional independence between its different symptoms. For instance, neglect may dissociate according to the sensory modality of stimulation, the spatial range of its influence or the spatial reference frame affected (see Vallar, 1998, for a review). Neglect may even be observed in mental imagery tasks, in which patients show a paucity of report for details on the left side of mentally evoked visual scenes (Bisiach and Luzzatti, 1978).

A second compelling reason for seeking to understand the neglect syndrome is with a view to its rehabilitation. Many patients recover from the acute manifestations of neglect relatively rapidly, but a significant proportion develop a chronic disorder (31.5% of patients with acute neglect still exhibit deficits at 3 months post-stroke: Cassidy et al., 1998). Several studies have found visual neglect to be a negative prognostic factor for functional outcome after stroke (e.g. Denes et al., 1982; Cherney et al., 2001). Moreover, the application of specific training regimes targeting neglect has been found to have concurrent

benefits for functional and motor recovery (Paolucci et al., 1996). Several sensory manipulations have now been shown to alleviate the symptoms of neglect transiently (see Vallar et al., 1997, for a review). However, longer lasting gains have proved elusive and many treatments that influence the clinical symptoms of neglect fail to generalise to everyday behaviours.

Recently, a new treatment for neglect was introduced by Rossetti et al. (1998). The procedure involves visuomotor adaptation to a rightward prismatic shift of the visual field (i.e. away from the neglected side). Rossetti et al. reported a reduction in neglect amongst six patients across a range of clinical measures. The treatment is remarkable for several reasons. First, large gains may follow a brief period of prism exposure (3-5 minutes). Second, the effects generalise widely across clinical measures of neglect, and subsequent studies have demonstrated that the effects may generalise to wheelchair navigation (Rossetti et al., 1999b), postural control (Tilikete et al., 2001) and neglect of mental imagery (Rode et al., 1999, 2001). Finally, the effects are persistent. Rossetti et al. (1998) measured benefits at two-hours post-adaptation which, if anything, were stronger than those observed immediately. More recently, three patients have been reported in whom benefits were observable after four days (Pisella et al., 2002). These observations suggest that visuomotor adaptation stimulates a reorganisation of the neural representation of space that develops autonomously after removal of the prisms, though the mechanisms of change remain obscure (see Farnè et al., 2002, for discussion of possible mechanisms).

Research into the prism adaptation treatment for neglect is in its infancy and many basic empirical questions present themselves. For instance, how generalised are the effects across tasks and across patients? What is the typical time course of the changes induced? Can cumulative benefits be achieved and how specific are the effects with respect to the various symptoms of neglect? We present a longitudinal case study of a patient (CS) with neglect of nine months' duration that may help shed some light upon these issues. We report that, despite the chronicity and severity of CS's neglect, a reliable influence of prism adaptation was obtained across a range of clinical tasks. We also provide a novel demonstration that the effects of prism adaptation are not limited to tasks with an explicit visual component, thus supporting the notion that neglect may affect higher levels of spatial representation. The results are discussed with regard to the rehabilitative potential of this procedure.

CASE REPORT

CS is a right-handed woman with nine years of formal education and professional secretarial qualifications. She is unmarried and had worked as a secretary until her retirement. At the age of 73, CS suffered a haemorrhagic stroke in the territory of the right middle cerebral artery. An MRI examination performed at two days post-stroke showed a large lesion in the right parietal lobe extending to postero-lateral occipital cortex, with sparing of medial occipital cortex. Her presenting clinical symptoms were left hemianaesthesia, left hemianopia to confrontation, left hemiplegia (with some movement in the upper limb), rightward cephalic deviation and severe left visual neglect. Formal assessment of CS's visual fields was performed by Humphrey perimetry at 14 months post-stroke. This examination confirmed that her left hemianopia was complete and homonymous.

An initial neuropsychological evaluation was performed at 7 days post-stroke at which time CS was alert and well oriented to time, place and person. Bedside testing found total extinction of left auditory and tactile stimuli and also of relatively leftward stimuli within the spared right visual field. At this time, formal assessments using visual materials proved impractical since, even when stimuli were placed on the extreme right side of her peripersonal space, CS would respond only to the rightmost part of the sheet. However, on three verbal sub-tests of the WAIS-R (information, vocabulary and digit span), CS performed adequately, achieving age-scaled scores of 10, 10 and 5 respectively (all within two standard deviations of the normal mean).

Formal assessment of CS's visual neglect was made at 9 weeks post-stroke, by which time she had recovered sufficiently to attend to visual material presented directly in front of her on a tabletop. CS was administered five standard screening tests for neglect: line cancellation (Albert, 1973), star cancellation (Wilson et al., 1987), scene copying (Gainotti et al., 1972), representational drawing (Wilson et al., 1987) and line bisection (see Materials and Methods for test details). Her performance is shown in Table I.

At six months post-stroke, now ambulatory, CS was discharged from hospital and initially attempted to make the transition to home living. However, her severe neglect proved a major obstacle to independent functioning, impeding many daily activities and causing her to have frequent collisions with objects on the left side. During the next month, she was transferred to a home for the elderly where the studies reported here were conducted. CS was 74 years old at the time of these studies.

EXPERIMENTAL STUDY

A longitudinal study was carried out in four sessions, spaced at weekly intervals, at approximately nine months post-stroke (39-42 weeks). In each session, a neglect test battery was administered. In order of administration, the battery comprised star cancellation, scene copying, line bisection, haptic circle centring and poem reading (see below for test details). In weeks 40, 41 and 42, CS was adapted to a rightward optical shift of 10° (see below for details). The event sequence for the four sessions was as follows:

Week 39: Pre-test¹

Week 40: Pre-test → prism adaptation → 2 hour post-test

Week 41: Pre-test → prism adaptation → immediate post-test²

Week 42: Pre-test → prism adaptation → 2 hour post-test

TABLE I
CS' Neglect Screening Performance at 9 Weeks Post-Stroke

Line cancellation L lines omitted R lines omitted	Star cancellation cancellation L stars omitted R stars omitted	Scene copying (5 items) Items copied (symmetrical items)	Drawing (3 items) Symmetrical items	Line bisection 10 × 20cm lines Mean error (SD)
100% 0%	100% 41%	2 (1) see Figure 2b	3	+ 59.8 mm (17.4)

¹Due to an oversight, the scene copying task was not performed at week 39.

²The planned two-hour post-test in week 41 had to be cancelled. An immediate post test was instead performed with a reduced battery that omitted the most time consuming test (haptic circle centring). To compensate for this disruption, a final session was added at week 42 with the event sequence matched to that of week 40.

MATERIALS AND METHODS

Test Details

All tasks were untimed and all stimuli were presented at body midline.

Star cancellation was taken from the Behavioural Inattention Test (Wilson et al., 1987). The A4 stimulus sheet contained 56 targets (small stars) pseudo-randomly interspersed with distracter items. The targets actually fell into six columns, with two additional targets located centrally. The experimenter indicated clearly the full extent of the sheet and crossed out the two central targets. CS was then asked to cancel the remaining small stars. The number of targets omitted in each lateral half of the sheet was counted.

Scene copying was adapted from Gainotti et al. (1972). CS was asked to copy a simple scene made up of five items (see Figure 2). Performance was scored in two ways: the number of items copied and the number of items symmetrically depicted.

In the *line bisection* task, CS was presented with 10 black horizontal lines, each 20 cm long and 2 mm thick and each centred on an A4 sheet of white paper. Rightward errors were scored as positive values and leftward errors as negative.

The *haptic circle centring* task employed a 30 cm diameter circular groove cut centrally into a 1 cm thick square of transparent perspex (37.5 × 37.5 cm). CS was blindfolded and the stimulus was placed in front of her with its centre at a radial depth of about 25 cm from her body. She was invited to explore the stimulus using her right hand, and was given up to five practice trials before each trial block. In each experimental trial, CS's right index finger was placed in the circumference groove at one of four points: North, East, South or West with respect to CS (she was not informed that these four points were used). She was then required to make one full (clockwise or anticlockwise) exploration of the circumference groove. While she did this, the experimenter placed a finger at the original starting point so as to block further movement of CS's finger when the circumference had been fully explored. When CS felt her exploration blocked in this way, she was asked to move her fingertip to the centre of the circle and then to bring it to rest. The experimenter recorded (to the nearest 5 mm) the lateral and radial coordinates of the response from a scale attached to the underside of the stimulus. Rightward and distal errors were scored with positive values; leftward and proximal errors were scored with negative values. Each time the task was administered, CS performed 48 trials divided into two blocks with a short break between blocks. Within each block, start position (North, East, South, West) and direction of exploration (clockwise, anticlockwise) were counterbalanced. At no time was CS allowed to see the stimulus circle.

The above task was designed as an approximate haptic analogue of the visual line bisection task. Previously, the tactile line bisection task has been used for this purpose in the study of neglect patients and normals. However, studies of unilaterally brain damaged patients have failed to detect any evidence of neglect phenomena using the tactile line bisection task (Fujii et al., 1991; Hjaltason et al., 1993; McIntosh, 1999). This insensitivity of tactile line bisection to neglect may be due partly to the adoption of counting strategies by subjects performing this task, a tendency that has been noted by several authors (Hatta and Yamamoto, 1986; Levander et al., 1993; McIntosh, 1999). The haptic circle centring task is a bi-dimensional bisection task that precludes the use of counting strategies and thus promotes the adoption of spatial strategies. In support of this claim, significant left neglect (abnormally large rightward errors) has been observed on this task amongst neglect patients in whom tactile line bisection failed to identify any significant lateral biases (McIntosh, 1999).

Finally, a *poem reading* task was administered. CS was presented with eight stanzas (32 lines) of "The Hunting of the Snark" by Lewis Carroll (1939), a poem with which she was unfamiliar. The choice of a poem that features occasional "portmanteau" words (i.e. pronounceable nonwords) was incidental, although it was anticipated that the presence of such nonwords might make CS's reading errors less apparent to her. The text was presented in 16 point arial bold lowercase font and was justified centrally. The poem was presented on a laptop screen in the first session and on A4 paper in subsequent sessions. A different section of the poem was presented each time (mean total of 256 words). CS was asked to read the poem aloud.

Following the division of neglect dyslexic errors advocated by Young et al. (1991), errors were scored separately for whole words omitted and for words that were attempted but misread. Conservative criteria were applied to ensure that errors that were not a consequence of left neglect were not counted. For errors of word omission, the criterion adopted was that the entire word was omitted and that no word lying to the left of the omitted word on the same line of text was attempted. Misreading errors for attempted words were determined according to the criterion introduced by Ellis et al. (1987): that the target and error words were identical to the right of an identifiable “neglect point” in each word, but had no letters in common to the left of their neglect points. For each administration of the poem reading test, the number of neglect word omissions was expressed as a percentage of the total number of words presented. The number of neglect misreading errors was expressed as a percentage of the total number of words attempted.

Prism Adaptation Procedure³

The adaptation procedure followed the method of Rossetti et al. (1998). CS wore a pair of goggles fitted with wide-field point-to-point prismatic lenses creating a rightward optical shift of 10°. Exposure consisted of 50 fast pointing movements made to visual targets presented 10° to left and right of the body midline, with 25 movements to each in pseudo-random order. A shelf was held under CS’s chin to prevent her viewing her hand at its starting position, but allowing her to see the targets and her terminal errors. The exposure period lasted for between three and five minutes.

RESULTS

For all of the following tasks, the most pertinent comparisons are between pre- and post-prism performance in the two matched sessions, conducted at weeks 40 and 42. For completeness, the immediate post-adaptation results from week 41 are also included in Figures 1-5.

Star Cancellation

Figure 1 shows the percentage of total targets omitted at each administration of the star cancellation test (black dots indicate omissions in the right half of the sheet). The overall effect of prism adaptation was statistically evaluated by pooling the pre-prism data across weeks 40 and 42 and similarly pooling the post-prism data. A Mann-Whitney U test confirmed that a significantly greater number of targets were cancelled following prism adaptation ($z = 2.77$, $p < 0.001$). Similarly, a significant effect of week was confirmed by comparing the pooled pre- and post-prism data of week 40 with those of week 42 ($z = 3.64$, $p < 0.001$).

Scene Copying

Figure 2 illustrates CS’s performance of the scene copying test. No example from week 39 is available (see Materials and Methods). However, CS had attempted the task four times previously and all of her copies are shown here,

³An anonymous referee has pointed out that the pre-prism assessments should ideally have been preceded by a “sham adaptation” procedure, in which the above pointing task is performed with no prismatic deviation. This modification, which would control for any beneficial effects of the pointing component of the adaptation procedure, is recommended for future case studies.

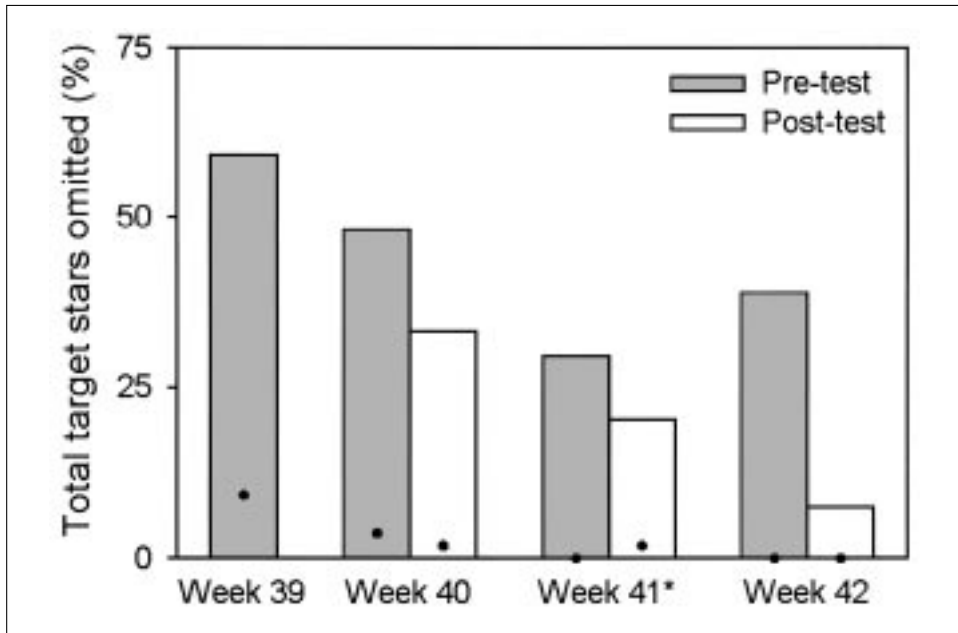


Fig. 1 – Percentage of total targets omitted at each session (black dots indicate omissions in the right half of the sheet). A clear improvement of performance is seen after prism adaptation. (*The post-test at week 41 was immediate, rather than two hours post-adaptation.)

illustrating the stability of her performance over this period. A dramatic improvement was stimulated by the first period of prism adaptation (week 40). This was largely maintained across sessions, even though novel stimuli were presented in subsequent weeks.

Line Bisection

Figure 3 shows CS's mean directional error at each administration of the line bisection test. The raw data from the matched sessions at weeks 40 and 42 failed to pass Levene's test for homogeneity of error variances [$F(3, 36) = 3.54, p < 0.05$]. Homogeneity of variance was obtained by adding a constant (+ 5) to the raw values to render them universally positive and squaring the result. The transformed data were entered into a two-way ANOVA by week (40, 42) and prism (pre, post). The main effect of prism was highly significant [$F(1, 36) = 22.48, p < 0.001$] and the effect of week narrowly reached significance [$F(1, 36) = 4.11, p = 0.05$].

Haptic Circle Centring

Figure 4 shows CS's mean lateral and radial circle centring errors. For lateral and radial errors separately, two-way ANOVAs by week (40, 42) and prism (pre, post) were conducted. For lateral errors, there was a highly significant effect of prism [$F(1, 188) = 8.58, p < 0.005$]. For radial errors, there were no significant effects.

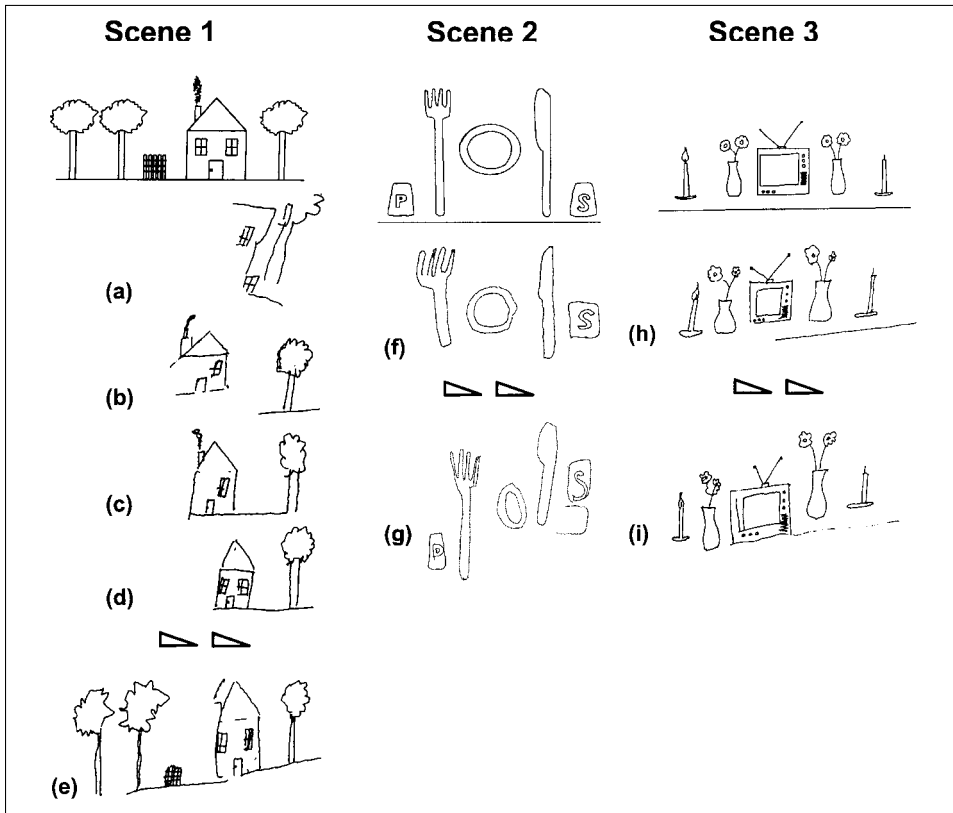


Fig. 2 – CS's scene copying performance at nine time points post-stroke with periods of prism exposure indicated. The three stimulus scenes employed are shown in the top row, with CS's copies below: (a) Week 1 (with the page presented in right hemisphere); (b) Week 9; (c) Week 22; (d) Week 40 (pre-test); (e) Week 40 (two hour post-test); (f) Week 41 (pre-test); (g) Week 41 (immediate post-test) (h) Week 42 (pre-test); (i) Week 42 (two hour post-test). Pre-adaptation performance was stable but a dramatic improvement was stimulated by the first period of prism adaptation. This was largely maintained in subsequent sessions even though novel stimuli were presented.

Poem Reading

Figure 5 shows CS's neglect dyslexic errors at each administration of the poem reading test. CS's neglect dyslexia was relatively mild and expressed primarily as whole word omissions, with 8.0% of words being omitted overall. Neglect misreading errors (shown as black dots in Figure 5) were produced on only 1.7% of attempted words overall. Prism adaptation had no consistent effect on CS's neglect dyslexia for either class of error.

DISCUSSION

The present case study, though necessarily limited in scope, provides several novel findings. First, it is clear that a prism adaptation treatment of five minutes or less can substantially improve the symptoms of neglect even in a severe case of

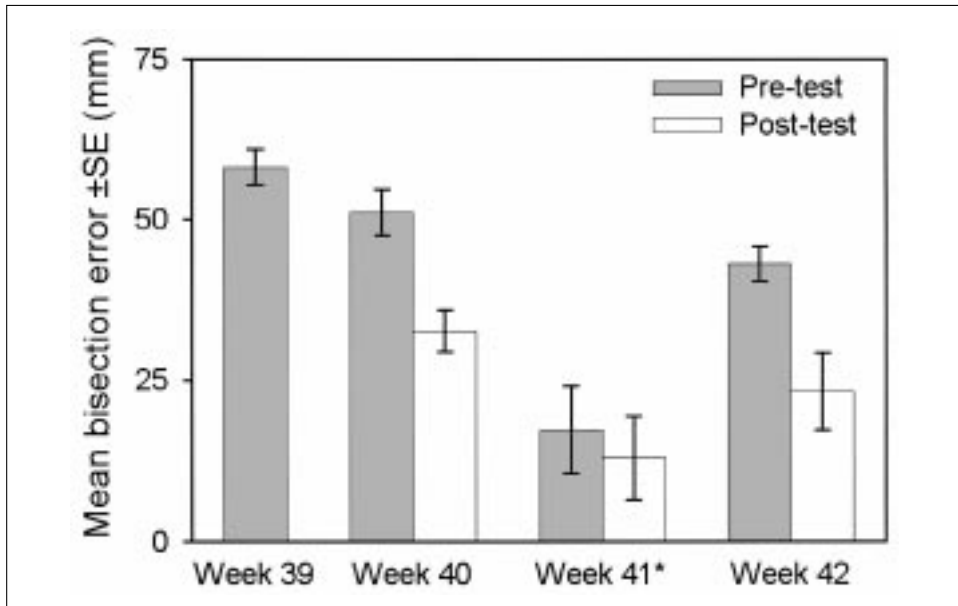


Fig. 3 – CS's mean bisection errors (\pm S.E.) at each session. A reliable reduction of rightward errors occurred following prism adaptation in weeks 40 and 42. (*The post-test at week 41 was immediate, rather than two hours post-adaptation.)

nine months' chronicity. The immediacy, strength and generality of CS's improvement suggest that there may be no upper limit to the time after stroke at which prism adaptation can be effective. In support of this, it may be noted that the group study of Rossetti et al. (1998) included patients up to 14 months post-stroke.

A second striking aspect of the present results is that prism adaptation induced a clear improvement of neglect on a spatial task with no explicit visual component. The haptic circle centring task was designed as a two-dimensional analogue of visual line bisection. However, unlike in the visual task, the haptic exploration of the circular stimulus was standardised to ensure that the circumference of the circle was fully explored before the midpoint judgement was made. Thus, the improvement of neglect observed on this task cannot be attributed to an improvement of haptic exploration. It is unclear whether the reduction of rightward errors was due to an alteration of CS's ability to mentally represent (or mentally explore a representation of) the stimulus circle, or to a reduction of a rightward manual response bias. The former possibility, however, is fully consistent with the findings that prism adaptation improves neglect on imagery tasks with no manual component (Rode et al., 1999, 2001). These cross-modal effects strongly support the view that prism adaptation induces changes not only in visuomotor coordination but also at higher cognitive levels of spatial representation (Rossetti et al., 1998, 1999b; Rode et al., 1999, 2001).

The generality of the influence of adaptation across tasks and sensory modalities suggests a recalibration of a high-level representation of space, and signals great promise for the therapeutic use of the prism treatment. Crucially, the benefits seem to generalise to activities and spatial scales directly relevant to

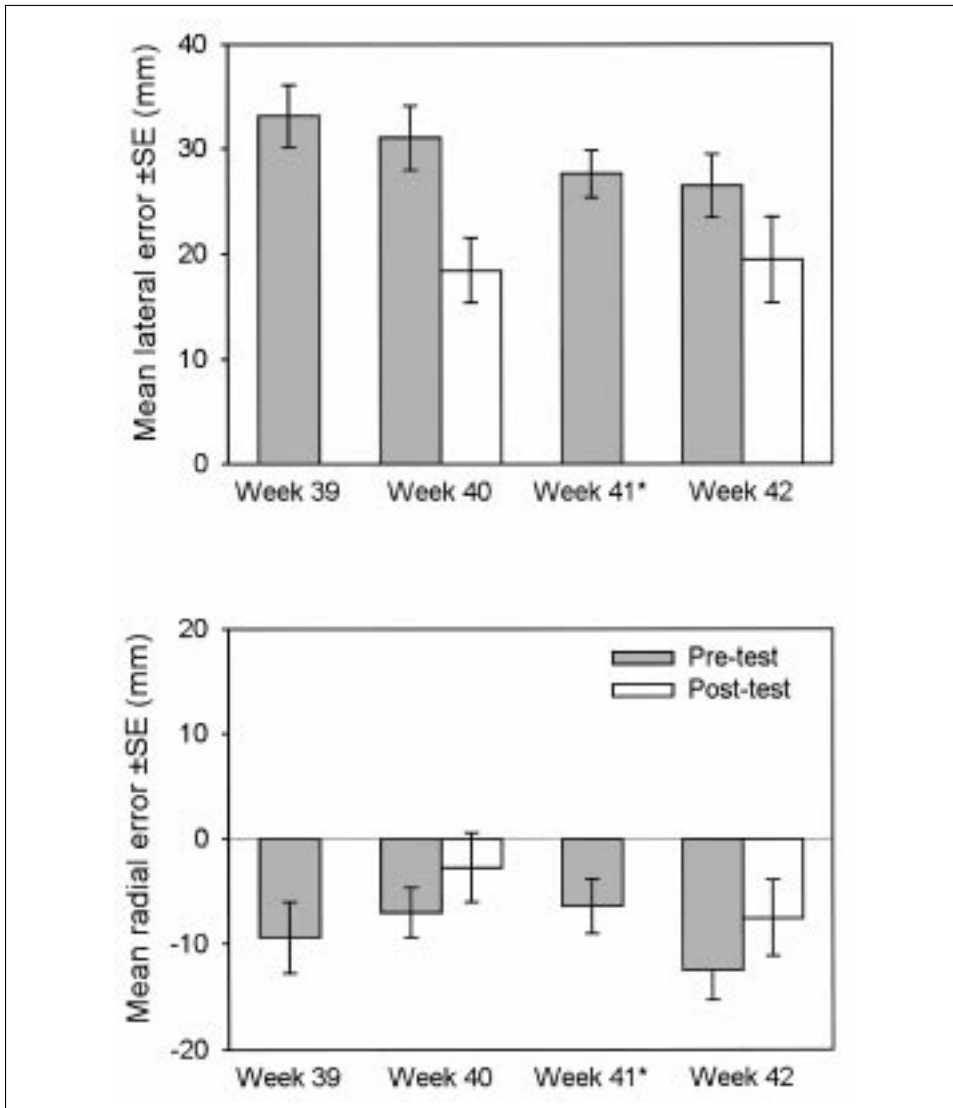


Fig. 4 – The upper graph shows CS's mean lateral circle centring errors (\pm S.E.) at each session. A reliable reduction in rightward errors occurred after prism adaptation in weeks 40 and 42. The lower graph shows CS's mean radial errors (\pm S.E.), with no reliable influence of prism adaptation. (*No post-test was performed for this task at week 41.)

real world settings. For instance, adaptation has positive effects on the ability of wheelchair-bound neglect patients to navigate around a hospital (Rossetti et al., 1999a). In the case of CS, there is anecdotal evidence that similar daily benefits were obtained. At the time that the present study commenced, nursing home staff would not allow CS to walk unaccompanied around the building due to the frequency with which she collided with obstacles on her left side (e.g. door frames, furniture). By week 42, the perceived danger of such collisions had

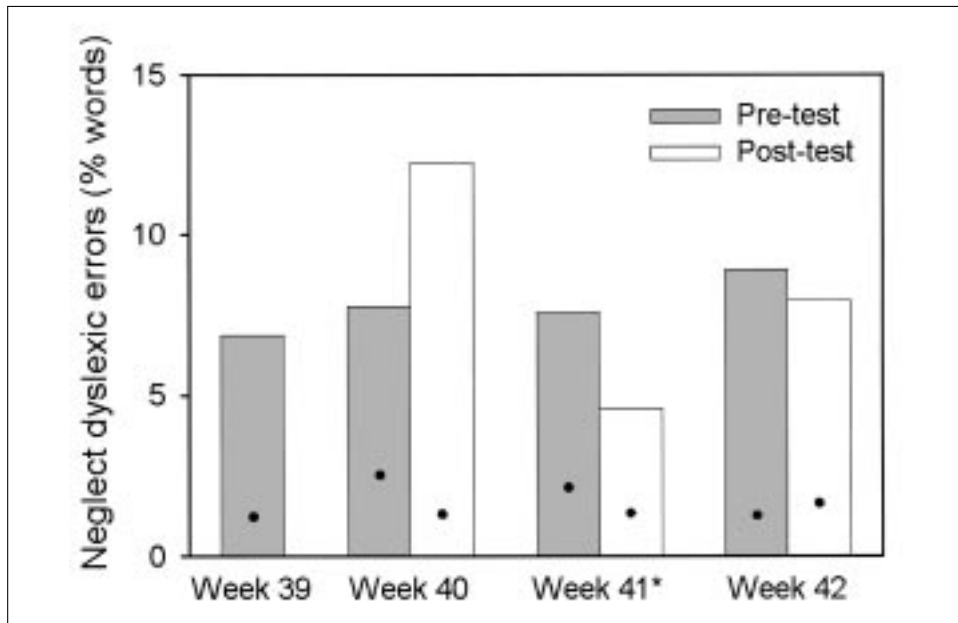


Fig. 5 – CS's neglect dyslexic errors at each session. Bars indicate word omissions (black dots indicate misreadings). There was no consistent influence of prism adaptation on either class of error. (*The post-test at week 41 was immediate, rather than two hours post-adaptation.)

reduced to the point that CS was now allowed to walk around the building alone. Unfortunately, when CS was visited again ten weeks later, she had returned to her former accident-prone status, indicating that the gains observed were not permanent.

In light of the generality of the effects of prism adaptation, it is surprising that CS's reading errors were apparently unaffected. It is tempting to relate this result to previous reports of dissociations of neglect dyslexia from other clinical symptoms of neglect (see Ellis et al., 1993). However, CS's performance should not be too readily interpreted in this manner. First, the variability of performance apparent in Figure 5 is rather high relative to the mild level of neglect dyslexia exhibited overall. It is possible that subtle effects of the prism treatment might have been masked by this background noise. Second, a recent group study of single word reading in neglect has demonstrated robust improvements in response to prism adaptation, indicating that neglect dyslexic misreading errors are not generally resistant to this intervention (Farnè et al., 2002). Similarly, text reading was among the tests that were performed better following prism exposure in the original group study of Rossetti et al. (1998). It is worth noting, however, that Rossetti et al did not distinguish between neglect dyslexic errors of word omissions and errors of misreading of single words. Given evidence that these classes of neglect dyslexic errors may have different underlying causes (Costello and Warrington, 1987; Young et al., 1991), it remains possible that they may be differentially susceptible to prismatic intervention.

Finally, it is noted that CS's data are consistent with a persistent influence of

prism exposure. The retention of gains over time was most obvious for the scene copying task, which CS performed almost perfectly on each occasion following the first prism treatment, even though novel stimuli were presented (the scene presented in week 42 was particularly intricate). Significant reductions of neglect across sessions were also found for line bisection and star cancellation, and there was a qualitative improvement across sessions for the haptic task. It must be conceded, however, that these effects are difficult to ascribe unambiguously to treatment effects, as distinct from possible effects of task repetition or spontaneous fluctuations of neglect severity. This caveat applies particularly to the star cancellation, line bisection and haptic circle centring tasks, as only two pre-treatment baseline measurements were obtained for these tasks (weeks 39 and 40) and qualitative improvements between baseline measurements were seen in all cases. Accordingly, whilst the present data are consistent with the conclusion that a single period of prism adaptation may yield measurable benefits at one week, this conclusion must be tentative. Moreover, whilst previous studies have found beneficial effects on line bisection up to four days after adaptation in individual neglect patients (Pisella et al., 2002; Rossetti et al., 1999a), a recent group study found no evidence of gains on a single-word reading task at one week post-adaptation (Farnè et al., 2002). Clearly, the typical time-course of the effects of prism adaptation in neglect, and its possible task-dependency, are topics requiring detailed further study.

CONCLUSIONS

This case study confirms the ameliorative power of the prism adaptation treatment for neglect, even when the disorder is severe and chronic. These effects are temporary, but remarkably long-lived in relation to the brevity of the prism exposure. The effects also generalise widely across tasks and the present study provides a first demonstration that prism adaptation can have an impact on haptic spatial judgements. This supports the view that the treatment stimulates high-level changes at the level of spatial representation rather than being limited to low-level sensorimotor re-mappings. It remains to be seen whether prism adaptation is beneficial for all varieties of neglect, and whether more intensive schedules of exposure can induce more permanent changes in spatial awareness. In particular, it will be of paramount importance to determine whether prism adaptation, in tandem with other therapies, can improve long-term outcome for patients with neglect.

Acknowledgements. We wish to thank CS for her patience and humour, all staff at the nursing home for their co-operation and Richard Weir for artistic assistance. This work was supported by grants from the Wellcome Trust (048060) and the Leverhulme Trust (F/00128/C) to ADM, and grants from the Wellcome Trust (010343) and INSERM (4P012E) to YR.

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