Sensorimotor incongruence exacerbates symptoms in patients with chronic whiplash associated disorders: an experimental study

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Abstract

Objectives. Incongruence between sensory feedback and motor output may serve as an ongoing source of nociception inside the CNS, and hence may contribute to the development of chronic whiplash associated disorder (WAD). It has been demonstrated that sensorimotor incongruence exacerbates symptoms and provokes additional sensations in patients with chronic pain. This study aimed to evaluate whether a visually mediated incongruence between motor output and sensory input aggravates symptoms and triggers additional sensations in patients with chronic WAD.

Methods. Thirty-five patients with chronic WAD and 31 healthy controls were subjected to a coordination test. They performed congruent and incongruent arm movements while viewing a whiteboard or mirror.

Results. All patients with chronic WAD (n = 35) reported sensory changes such as increased pain, tightness, loss of control, dizziness or feelings of peculiarity at some stage of the test protocol. No significant differences in frequency and intensity of sensory changes were found between the various test stages (P > 0.05). In the healthy control group, 18 (58%) subjects reported sensory changes at some stage of the test protocol, with the highest number during the incongruent mirror stage (n = 17), corresponding to the highest level of sensorimotor incongruence. The pattern of reported sensory changes during the congruent and incongruent stages was significantly different between both groups (P < 0.05).

Conclusion. This study demonstrates an exacerbation of symptoms and/or additional sensory changes due to reducing or disturbing the visual input during action, indicating altered sensorimotor central nervous processing and altered perception of distorted visual feedback in chronic WAD.

Key words: sensorimotor incongruence, chronic WAD, sensory changes, pain, chronic pain, altered sensorimotor central nervous processing.

Introduction

Chronic whiplash associated disorders (WAD) occur in 10–50% of people involved in an acute whiplash trauma [1–4]. The underlying mechanism and clinical pattern of chronic WAD are complex. Previous research demonstrated sensorimotor dysfunctions such as impaired movement control of the head and neck [5–9]. However, these cervical dysfunctions appear to be non-specific for chronic WAD and their prognostic value seems rather limited [10].

Recent scientific research stresses the involvement of the CNS. There is consistent evidence for central hypersensitivity in chronic WAD [11, 12]. Data regarding neuronal plasticity are available [13, 14]. The regional cerebral blood flow in the somatosensory cortex, medial frontal, posterior cingulate and parahippocampal gyrus were found to be increased in chronic WAD in the resting state [14]. Adaptive grey matter changes were observed...
Sensorimotor incongruence in chronic WAD

in the anterior cingulate and dorsolateral prefrontal cortex, thalamus and cerebellum in a group of WAD patients with chronic post-traumatic headache [13]. The net result may be fusion and/or overlapping of the receptive fields, which in turn may disrupt the body schema.

Cortical reorganization and disruption of the body schema, as seen in patients with chronic pain such as phantom limb pain, chronic back pain (cLBP) and those with complex regional pain syndrome (CRPS) [15–17], have been associated with sensorimotor incongruence and the development of pain [18]. In the same way as a conflict between visual and vestibular input results in motion sickness, incongruence between sensory feedback and motor output may lead to the experience of sensations [18].

McCabe and colleagues investigated this cortical pain model. They demonstrated that sensorimotor incongruence, mediated by incongruent visual input, triggers sensations in healthy people and aggravates pain in patients with FM [19, 20]. Likewise, we recently found that an experimentally induced sensorimotor conflict provokes sensations in asymptomatic, healthy violinists, whereas it exacerbates symptoms in violinists with unexplained musculoskeletal disorders [21]. Others have demonstrated that a conflict provoked by manipulating the proprioceptive input induced feelings of peculiarity, swelling and foreignness, but not pain, in healthy people [22]. The sensations may be warning signals, produced by the CNS, alerting the subject to incongruence between sensory feedback and motor output [23].

Given the impaired sensorimotor control and the indication for cortical plasticity in chronic WAD, incongruence between motor output and sensory feedback is likely to occur. No studies addressing the role of sensorimotor incongruence in chronic WAD are currently available.

The aim of this study is to evaluate whether a visually mediated incongruence between sensory input and motor output increases symptoms and triggers additional sensations in patients with chronic WAD. We hypothesize that patients with chronic WAD are more susceptible to visually mediated changes between sensory feedback and motor output than controls.

Methods

Subjects

Participants were recruited through an advertisement on the World Wide Web and from the medical database of the local Red Cross medical care unit. The inclusion criteria were experiencing chronic symptoms resulting from a whiplash trauma and fulfilling diagnostic criteria of WAD I–III as defined by the Quebec Task Force classification [3]. Chronicity was defined as complaints persisting for at least 3 months. Subjects were excluded if they were classified as WAD IV [3].

Healthy control subjects were recruited from the university college staff, and family members and acquaintances of the researchers. Controls were not allowed to participate if they ever had experienced a whiplash trauma, suffered from (persistent) pain or neck–shoulder–arm symptoms, or sought medical help for neck–shoulder–arm symptoms in the past 6 months.

Subjects were asked to discontinue analgesic and anti-inflammatory drugs 48 h before testing. This duration was chosen based on ethical considerations and the fact that analgesic effects are mostly limited in time [24]. Subjects were instructed to avoid physical exertion and refrain from consuming nicotine, alcohol and caffeine 24 h before testing. Subjects were excluded if they were pregnant or if they suffered from any cardiovascular or neurological disease.

The sample size was calculated based on the findings of previous research. It has been demonstrated that 89% of the patients with chronic pain and 58% of the healthy subjects reported sensations when performing a task simulating sensorimotor incongruence [19, 20, 22]. Considering these results, proportions of 0.89 and 0.58 were used. An a priori power analysis determined that at least 30 subjects per group were required, with a power of 0.80 and \( \alpha \leq 0.05 \). The control and chronic WAD groups were age and gender matched.

Procedure

Before study participation, subjects carefully read an information leaflet. In the leaflet, the participants were informed that the experiment consists of a coordination task involving both arms. No further explanation concerning the task was given and no association with pain and other sensations was made. Written informed consent was obtained from all participants before testing in accordance with the Declaration of Helsinki. The study was approved by the Human Research Ethics Committee of the Antwerp University Hospital. A standardized questionnaire was used to collect personal characteristics, and accident- and health-related information. The WAD group filled in the Neck Disability Index (NDI). Next, they were subjected to a sensorimotor incongruence coordination task.

Measurements

Self-reported questionnaire

The NDI was used to evaluate participants’ neck pain and disability (score out of 100). The NDI was found to be reliable and valid [25–28].

Bimanual coordination test

The assessment apparatus and procedure employed in the current study have been described in detail by McCabe et al. [19, 20] and were used in our experimental study in professional violinists [21]. The method is briefly explained here. For details, the reader is referred to the previous studies [19–21]. The assessment apparatus, consisting of a mirror (intervention side) and a whiteboard (control side), was positioned on the subject’s anterior midline (Fig. 1). The subjects were asked to flex/extend both arms in a congruent/incongruent manner while viewing the whiteboard/mirror and attending to a reference point (a horizontal line at the level of the subject’s
At the end of each stage, two open-ended questions were asked: ‘How did it feel?’, ‘Were you aware of any changes in either limb?’. To prevent introducing bias, no further explanation was given and no suggestions towards sensations were made. In the case of reported sensations, the intensity was rated on a numerical rating scale from 0 to 10 [29]. If sensations were reported, a rest period was included until the symptoms had disappeared or returned to their baseline status.

Experiencing pain at the time of testing is likely to introduce bias into the study. Therefore control conditions (i.e. flex/extend both arms in a congruent/incongruent manner without viewing the whiteboard/mirror) were included and patients were compared with controls. All the limb assessments were executed consecutively and in random order. The abbreviations for the test and control stages (CW: congruent whiteboard; IW: incongruent whiteboard; CM: congruent mirror; IM: incongruent mirror; CC: congruent control; IC: incongruent control) will be further used in this article.

Statistical analysis
Statistical analyses were conducted using SPSS 18.0 for Windows (SPSS Inc., Chicago, IL, USA). Normality of the variables was tested with the Kolmogorov–Smirnov test. Comparability of the groups for gender distribution, education level and age was verified with Pearson’s $\chi^2$-test and independent-sample t-test. A McNemar test (binomial distribution) was used to compare the frequencies of the experienced sensations within the congruent (i.e. CW and CM) and incongruent test stages (i.e. IW and IM) and between the test and control stages in either group. A paired samples t-test and a Wilcoxon signed rank test were used to compare the intensities of sensations across the various stages in the WAD and control group, respectively. Two repeated-measures analyses of variance (ANOVA) were conducted to evaluate the pattern of sensations across the control, whiteboard and mirror condition within each group and between both groups while performing congruent and incongruent movements, respectively. Each model had one between-subjects factor group (chronic WAD, controls) and one within-subjects factor stage (control, whiteboard, mirror). If no sensations were reported during an individual stage, the frequency and intensity score was counted as 0 and that score was recorded for statistical analysis. The significance level was set at 0.05.

Results

Group characteristics
Thirty-five patients with chronic WAD (26 women and 9 men) and 31 controls (24 women and 7 men) volunteered for the study. The mean (s.d.) age was 43.8 (9.58) and 43.19 (16.11) years for the WAD and control groups, respectively. Both groups were comparable for age, gender distribution and education level ($P > 0.05$). The mean (s.d.) score of the NDI was 44.36 (12.64) in the chronic WAD group. One subject reported mild pain and disability (NDI scores between 10 and 28), and 34 subjects were classified as having moderate/severe pain and disability (NDI score $> 30$).

Reported sensory changes in response to the various stages

Patients with chronic WAD
During the performance of the test protocol, all patients ($n = 35$) reported sensations at some stage in addition to or increased compared with those experienced at baseline. The frequencies of reported sensations across the
various test stages are presented in Table 1. No significant
differences in frequencies were found between the various
test stages in the WAD group, neither between the con-
gruent nor between the incongruent test stages ($P > 0.05$; Table 1).

The reported sensations were described as pain (60%),
tightness (37%), loss of control (34%), dizziness (31%),
feelings of peculiarity (29%), discomfort (20%), weight
changes (20%), tiredness (17%), tingling (11%), temperature
changes (9%) and perceived loss/additional arm
(9%).

In cases of reported sensations, the mean intensities
are presented in Table 2. No significant differences in
intensities of reported sensations were found between
the various test stages ($P > 0.05$; Table 2). The sensations
disappeared/returned to baseline level when visual input
was restored.

During the control stages, 27 (77%) patients reported
pain, discomfort and/or tightness (Table 1). The mean
intensities of reported sensations are presented in the
right columns of Table 2.

Significantly more sensations were reported during the
CW, IW and IM stages compared with the control stages
($P < 0.05$; data not shown). No significant difference in frequency
was observed between the CC and CM stages
($P = 0.070$; data not shown). The mean intensities of re-
ported sensations were significantly different between the
control and test stages ($P < 0.05$; data not shown).

Healthy controls
In the control group, 18 (58%) subjects reported sen-
sations at some stage of the test protocol, with the
highest number ($n = 17$) during the IM stage (i.e. the
stage with the highest level of sensorimotor incongru-
ence; $P = 0.003$; Table 1). The frequencies of reported
sensations across the test stages are shown in the
lower row of Table 1. No significant difference in fre-
quency was observed between the congruent test
stages ($P > 0.05$; Table 1). Mostly, the sensations were
reported in the hidden arm and disappeared when visual input was restored.

The controls reported sensations such as loss of control
(32%), feelings of peculiarity (29%), weight changes
(10%), discomfort (6%) and perceived loss/additional arm (3%). In cases of discomfort, uncomfortable and la-
borigious feelings were reported by the controls. None of
them reported pain.

The intensities of the reported sensations are presented
in the lower rows of Table 2. The intensity was significantly
higher during the IM stage compared with the IW stage
($P = 0.002$; Table 2). No significant difference in intensity
was found between the congruent test stages ($P > 0.05$; Table 2).
None of the controls reported sensations during the
control stages. Significantly more sensations were re-
ported during the IM and IW stages compared with the
IC stage ($P = 0.002$; Table 2). No significant difference in intensity
was found between the congruent test stages ($P = 0.070$; data not shown).

The intensity of reported sensations was significantly higher
during the IM stage compared with the IC stage
($P < 0.05$; data not shown). No significant differences in
intensities were observed between the control and the
other test stages ($P > 0.05$; data not shown).

### Table 1 Frequency of reported sensory changes per stage of the coordination test in patients with chronic WAD ($n = 35$) and healthy controls ($n = 31$)

<table>
<thead>
<tr>
<th>Group</th>
<th>CW, %</th>
<th>CM, %</th>
<th>P</th>
<th>IW, %</th>
<th>IM, %</th>
<th>P</th>
<th>CC, %</th>
<th>IC, %</th>
</tr>
</thead>
<tbody>
<tr>
<td>cWAD</td>
<td>33 (94)</td>
<td>33 (94)</td>
<td>1.000</td>
<td>34 (94)</td>
<td>33 (94)</td>
<td>1.000</td>
<td>27 (77)</td>
<td>27 (77)</td>
</tr>
<tr>
<td>HControl</td>
<td>4 (13 )</td>
<td>4 (13 )</td>
<td>1.000</td>
<td>6 (19 )</td>
<td>17 (55)</td>
<td>0.003</td>
<td>0 (0)</td>
<td>0 (0)</td>
</tr>
</tbody>
</table>

Significant at $P < 0.05$. cWAD: chronic whiplash associated disorders; HControl: healthy controls.

### Table 2 Mean and median intensities of reported sensory changes per stage of the coordination test in patients with chronic WAD ($n = 35$) and healthy controls ($n = 31$)

<table>
<thead>
<tr>
<th>Group</th>
<th>CW</th>
<th>CM</th>
<th>P</th>
<th>IW</th>
<th>IM</th>
<th>P</th>
<th>CC</th>
<th>IC</th>
</tr>
</thead>
<tbody>
<tr>
<td>cWAD</td>
<td>4.69 (2.22)</td>
<td>4.86 (2.13)</td>
<td>0.447</td>
<td>4.71 (2.24)</td>
<td>5.11 (2.19)</td>
<td>0.210</td>
<td>2.71 (1.89)</td>
<td>2.71 (2.11)</td>
</tr>
<tr>
<td>HControl</td>
<td>0.35 (1.17)</td>
<td>0.19 (0.54)</td>
<td>0.581</td>
<td>0.58 (1.36)</td>
<td>1.61 (1.90)</td>
<td>0.002</td>
<td>0 (0)</td>
<td>0 (0)</td>
</tr>
</tbody>
</table>

Significant at level $P < 0.05$. cWAD: chronic whiplash associated disorders; HControl: healthy controls; IQR: interquartile range.
Pattern of sensations in patients with chronic WAD and controls

Congruent stages

Fig. 2 shows the pattern in intensity of sensations across the CC, CW and CM stages in both groups. The results reveal a significant stage effect ($F = 38.592$, df = 2, $P < 0.001$) and group effect ($F = 121.993$, df = 1, $P < 0.001$), and a significant stage × group interaction ($F = 23.161$, df = 2, $P < 0.001$).

Incongruent stages

Fig. 3 presents the pattern in intensity of sensations across the IC, IW and IM stages in both groups. The results indicate a significant stage effect ($F = 50.336$, df = 2, $P < 0.001$) and group effect ($F = 78.611$, df = 1, $P < 0.001$), and a significant stage × group interaction ($F = 6.154$, df = 2, $P = 0.003$).

Discussion

This study demonstrates for the first time the role of visually mediated changes between motor output and sensory feedback in chronic WAD. The findings revealed an exacerbation of symptoms and/or additional sensations due to reducing/disturbing the visual input during action in patients with chronic WAD.

Reported sensory changes in response to various stages

The brain predicts the motor output. Through input from the eyes, skin, joints and muscles, the CNS is continuously informed about the actual movements. Simultaneously, predicted and actual motor plans are compared. This is important for updating the current body schema and smooth execution of complex motor tasks. The prefrontal and parietal cortices are crucial areas involved in the integration and monitoring of complex motor actions [30–32].

In cases of incongruence between sensory feedback and motor output, the CNS produces sensations alerting the person to the conflicting information being processed [18, 19]. The results of this study confirmed this notion: 55% of the controls reported sensations when sensorimotor conflict was provoked by manipulating the visual input (i.e. during the IM stage). Due to reflection of the visualized arm in the mirror, the visual input was manipulated and conflicts with the proprioceptive feedback of the hidden arm. This is in concordance with the findings of McCabe et al. [19] in healthy subjects. They indicated that some healthy people are more susceptible to such conflict and therefore experienced more sensations than other non-susceptible subjects.

None of the controls reported pain during performance of the coordination test, which confirms the results of previous reports. Moseley et al. [22] found symptoms of peculiarity, foreignness and swelling, but not pain, in controls due to manipulating the proprioceptive, instead of the visual, input. However, different kinds of incongruence were induced. Moseley’s work used a method generating illusory motion of a body part, while in our study actions were maintained in the face of visually mediated incongruence. Static body schema perception involves mostly tactile and deep somatosensory signals, whereas movement-related signals derive from proprioceptive receptors in muscles and joints.

In contrast, sensations of pain (e.g. pins and needles, moderate aching and/or definite pain) were observed in one study [19]. Harris’s proposition that pain results from cortical changes in the sensorimotor cortex and sensorimotor incongruence [18] still needs to be corroborated. Prospective cohort studies are required to investigate the cause–effect relationship between pain and cortical plasticity. The warning signals may be experienced as pain when the sensorimotor incongruence persists to the extent that the individual (pain) threshold is reached.
This may explain why the controls in this study reported only sensations, and not pain.

In the WAD group, reducing/disturbing the visual feedback (via moving a moving arm) seems to be sufficient to exacerbate symptoms and/or elicit additional sensations. According to McCabe et al.’s findings in patients with FM [20], no significant differences in reported sensations were found between the different test stages. These results indicate altered sensorimotor central nervous processing and support the presumption of distorted body schema in chronic WAD.

Body schema has been described as the actual performance of the body in its environment, which involves an integration of proprioceptive, vestibular, somatosensory and visual input from the periphery that interrelate with motor system [33]. De Vignemont [34] described the body schema as ‘a cluster of sensorimotor representations that are action-oriented including bodily properties relevant for action programming, prediction and sensory feedback’. It has been proposed that there are at least two distinct types of body representation, the body schema and the body image [33, 35]. However, there is widespread confusion about these terms. Deficits of body schema and body image are often part of the same syndrome and hard to separate.

Disrupted body image has been demonstrated in chronic pain conditions such as CRPS and cLBP [36–38]. Moseley et al. [39] found that distorted visual feedback of a moving limb can modify the body image and modulate pain in CRPS. In that study, magnifying the visual image of a moving limb led to a significant pain increase, whereas minimizing the view decreased pain. It has been hypothesized that magnifying the image of a limb introduces conflict between vision and proprioception, whereas minimizing the image decreases the ownership of a limb, leading to pain reduction in that limb [39].

Since visual input influences the construction of the body schema, it has been assumed that disturbing the visual input can modulate the body schema and cause sensorimotor incongruence at the cortical level. The CNS produces warning signals if the extent of incongruence is large enough to reach the individual threshold. In cases of lowered threshold, likely to occur in the WAD group, the CNS will be alerted during the stages with lower visually mediated incongruence (i.e. CW, IW and CM stages) as well. Then, the body schema will be further modulated, leading to an exacerbation of pain and other symptoms, explaining the results in the WAD group. No further conclusions concerning the causal link between cortical reorganizations, distorted body schema and pain can be made. Caution is needed when interpreting and comparing these results with those observed in other chronic pain conditions. Further studies addressing cortical reorganization and disrupted body representation in chronic WAD are warranted.

The results indicate that patients with chronic WAD depend greatly on visual input of the neck and upper limbs to reassure the CNS about limb movements. Due to the observed cervical motor dysfunction [5–8], patients with chronic WAD may rely heavily on visual input of the cervical region. Impaired shoulder proprioception and reduced acuity of goal-directed arm movements were found in patients with chronic WAD [40, 41]. Indeed, impaired movement control and reduced performance accuracy, as documented in patients with chronic WAD, could be the result of body schema disruption producing incorrect reference information for motor planning.

This study confirmed the involvement of the CNS in chronic WAD. The coordination test included only arm movements; no neck movements were performed. Since the head and neck position were not standardized during the test protocol, minor head/neck movements might have occurred during the experiment. The observed exacerbation of symptoms and the experience of additional sensations due to moving the arms while viewing the mirror/whiteboard towards central processes, instead of peripheral, as underlying mechanism of chronic WAD.

The interaction between disrupted body schema and chronic pain seems to be bi-directional [42]. On the other hand, a distorted body schema may be the result of central sensitization. The wide dynamic-range neurons in the dorsal horn of the spinal cord have a dual function. First, they activate in response to nociceptive and weak mechanical stimuli. Second, they inform the brain of information arising from receptors in skin, joints, muscles and viscera relevant to the construction of the body schema. Nociception modifies the functioning of the wide dynamic-range neurons. Subsequently, repeated or sustained noxious stimulation may result in prolonged activity of the dorsal horn neurons leading to central sensitization [43]. Central sensitization involves altered sensory processing and increased activity in brain areas such as the insula, various brain stem nuclei, the anterior cingulate, prefrontal and parietal cortex [44]. As a result, the receptive fields expand and overlap each other, leading to body schema distortion and consequently exacerbation of symptoms. Widespread hypersensitivity, indicative of central sensitization, was documented in chronic WAD [45–48].

**Pattern of sensations in patients with chronic WAD and controls**

The evolution in intensity of sensations was significantly different between both groups during the congruent as well as the incongruent stages. The WAD and control group differ in time course of sensations across the congruent stage, with a rising curve for the WAD and a flat curve for the control group. In the WAD group, the strongest increase was found between the CC and CW stages, with no significant increase observed between the CC and CW stages.

Across the incongruent stages, the rising curve in intensity of sensations was also found in the control group. However, the location of increase was different between both groups. In the control group, the strongest increase was observed between the IW and IM stages, whereas in...
the WAD group the strongest increase was found between the IC and IW stages. These results indicate vulnerability to sensorimotor conflict in healthy ‘susceptible’ subjects. In patients with chronic WAD, reducing/disturbing the visual input during action was sufficient to exacerbate symptoms, but no additional effect was observed during the IM stage. Further modulation of disrupted cortical body representation in patients with chronic WAD could explain these findings.

In conclusion, patients with chronic WAD present an exacerbation of symptoms and additional sensations in response to visually mediated changes during action. This study indicates an altered perception of distorted visual feedback and supports the involvement of the CNS in chronic WAD.

### Rheumatology key messages

- Visually mediated changes between sensory feedback and motor output exacerbate symptoms in chronic WAD.
- Sensorimotor conflict can induce sensations in susceptible healthy subjects.
- These results support involvement of the CNS and suggest distorted body schema in chronic WAD.

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