Referred sensations in patients with complex regional pain syndrome type 1

C. S. McCabe, R. C. Haigh, P. W. Halligan* and D. R. Blake

Objectives. This study sought to explore and characterize referred sensations (RS) in patients with complex regional pain syndrome (CRPS) type 1 and test the hypothesis that pain in CRPS is associated with central sensory changes.

Methods. Subjects underwent standardized neurological examination involving light touch, pinprick and vibration sense with eyes closed and then with eyes open. The subjects described the location and sensation emanating from the stimulated site and whether they experienced any sensations (similar or different) elsewhere.

Results. Five of 16 subjects recruited demonstrated RS. These were experienced in real time, were modality specific (touch and pinprick) and were located on the body part immediately adjacent, on Penfield’s cortical homunculus, to the stimulated site. The RS were diminished or absent when the subject visualized the stimulated area. They disappeared when stimulation ceased and on clinical improvement.

Conclusions. This is the first report of RS in CRPS and provides further evidence of central reorganization in what was previously thought to be a peripheral disorder.

KEY WORDS: Complex regional pain syndrome type 1, Referred sensations, Central reorganization.

Referred sensations (RS) are somatosensory feelings that are perceived to emanate from a body part other than, but in association with, the body part being stimulated. They have been reported following limb amputation [1], somatosensory deafferentation [2], local anaesthesia [3], stroke [4] and spinal cord injury [5]. Collectively these studies have shown that the referred sites (the body part not physically touched) are non-random and often closely correspond to the body structure that is immediately adjacent to the stimulated site on the cortical topographical map first described by Penfield [6]. In the case of an amputated upper limb, patients report sensation in their phantom when parts of the face are lightly stroked [1]. These aberrant somatosensory, but reliable, sensations were interpreted as resulting from central sensory reorganization following disconnection or dysfunction of sensory pathways [1].

Sensory and motor disturbances occur in patients with complex regional pain syndrome (CRPS) [7]. This condition usually follows after a minor peripheral injury to a limb, but may occur spontaneously. It may be acute or chronic; type 1 is differentiated from type 2 by the absence of neural trauma [8]. The clinical features of CRPS are well described and can be divided into five main areas: pain, oedema, autonomic dysfunction, movement disorder and trophic changes [9]. Pain is the dominant feature with patients typically describing a burning sensation that occurs spontaneously or spreading beyond the site of injury. The pain may be intermittent or persistent and specific sensory changes may include, allodynia, hyperalgesia and hyperaesthesia. Hemisensory impairment with decreased temperature and pinprick sensation ipsilateral to the CRPS-affected limb has also been reported [10]. This may involve the CRPS-affected limb only or encompass a more extensive area on the same side of the body. Where the latter, more generalized sensory deficit occurs this has been shown to correlate significantly with longer disease duration and a higher report of mechanical allodynia/hyperalgesia [11]. It has been proposed that these changes are attributable to functional disturbances in the processing of noxious events in the thalamus and a hemisensory deficit is indicative of subcortical brain plasticity. What has not been described previously is the presence of sensation in...
another body part when the painful site, or an area distal to that site, is stimulated, i.e. referred sensations (RS).

CRPS pain shares many similar characteristics to amputee phantom limb pain, mislocalized, intense and burning. As neural plasticity occurs in a variety of pain syndromes [12, 13] and because of the nature of CRPS pain, we predicted that if the disturbed peripheral sensations in CRPS type 1 were associated with central sensory changes, then evidence of this would be found in some patients as referred sensations. Furthermore, we hypothesized that these referred sensations would be perceived to emanate from the body structures immediately adjacent to the stimulated site and in keeping with their topographical location on the Penfield homunculus as in phantom and allied pain states. We specifically selected those patients with CRPS type 1 as we wished to discover whether central reorganization occurs even where there is no evidence of local peripheral nerve damage.

We therefore set out to explore and characterize referred sensations in patients with CRPS type 1 and we present five case studies where referred sensations were found to be present. The Bath Local Research Ethics Committee granted ethical approval and informed patient consent was gained.

Method and participants

Participants

Subjects who conformed to the diagnostic criteria for CRPS type 1 [14] were recruited over a 2-yr period from the out-patient and in-patient departments of the Royal National Hospital for Rheumatic Diseases, Bath.

Methods

Subjects were assessed on initial presentation and weekly until either symptom resolution occurred or, in those with chronic disease, discharge from in-patient care. Each assessment took the following format.

Subjects were placed in a supine position with the head of the couch elevated so that they could view all their limbs. They were asked to close their eyes and describe to the researcher any sensations they were experiencing, first in their unaffected limbs and then their affected limb. This first stage was used to accustom the subjects to focusing upon themselves and to establish baseline descriptions for unaffected limbs. Where the upper limb was affected the subject was first questioned about their legs followed by their unaffected upper limb and finally the affected limb. Conversely when the lower limb was involved the upper limbs were described first.

All subjects then underwent a standardized neurological examination testing light touch, pinprick and vibration sense first with their eyes closed and then with their eyes open. All limbs, lower spine and face were examined and sham trials, combined with a random order, were employed to reduce the possibility of patient suggestibility. Each time the subject was touched they were asked to describe the location of the stimulated site, the sensation emanating from it and whether they experienced any sensations (similar or different) anywhere else.

Results

Over the 2-yr recruitment period, 16 subjects (13 female and three male) who met the entry criteria were recruited. Only five showed evidence of referred sensations and it is the findings of these five (four female and one male) that will be presented (Table 1). There was no difference in age, disease duration, levels of pain or severity of disease (Table 2) between those who presented with RS and those who did not.

The five subjects had a disease duration of 3 weeks to 6 yr (median 3 yr) and were aged from 24 to 57 yr (mean 36.8 yr). All had a single limb affected (two upper limb, three lower limb). In four cases (cases 1, 3, 4 and 5) the condition was spontaneous in onset and only in case 2...
did it occur following injury. All reported pain that extended beyond the originating site with associated allodynia, hyperalgesia and vasomotor changes. None of the subjects had ever reported any previous perception of referred sensations to their physician.

Case 1
A 28-yr-old woman was admitted for in-patient rehabilitation with a 3-week history of progressive pain in her left hand for which there was no obvious triggering event. Her initial symptoms, prior to the onset of pain, were that of mottling of the fingertips. An intense burning pain involving all four fingers, but excluding the thumb, rapidly followed. Cold and light touch aggravated the pain and feeling of swelling. The patient held her limb in a flexed and pronated position close to her chest. Her hand was cold to touch and quantitative thermal imaging identified a 2.0°C temperature difference between the right and left forearms with the left cooler. With her eyes closed she described her hand as excessively large ‘like a blow up hand’. This phantom sensation of swelling extended to the thumb, despite no perceived involvement of this digit in her pain description. The patient was aware that this sensation was disproportionate to the degree of swelling that she observed. When the tip of the third finger of the affected hand was touched with a cotton bud, still with eyes closed, she experienced a stroking sensation over the lower left jaw (Fig. 1a). This sensation was modality specific with the subject reporting a pinprick sensation on her lower left jaw when the same finger was touched with a needle. The referred sensation only occurred at the time that the third finger was touched and there was no residual effect once the researcher stopped. Vibration was not referred. When the subject’s left lower jaw was touched there were no reciprocal referred sensations experienced in the left hand.

The above examination was repeated with the subject looking at their hand as their affected limb was touched. Permitting direct visual feedback prevented the experience of referred sensations.

Over the next 2 weeks the referred sensations could be evoked at each assessment. However, by week 3 the subject no longer perceived her hand as swollen and referred sensations were lost. By 6 weeks all vasomotor changes were reversed and no pain was felt.

Case 2
A 34-yr-old woman presented to the out-patient department 8 weeks after an industrial accident, sustaining a minor injury to her left foot but no neural trauma. The initial pain of the injury settled, but returned 2 days later. On admission she described a stabbing pain from the toes to mid-calf. The foot was swollen, mottled in colour, hyperalgesic, allodynic and hyperhydrotic. The foot and calf were cold to the touch. Quantitative thermal imaging showed a 2.5°C difference between the right and left foot. With her eyes closed the subject noted that her left foot appeared enlarged, greater in size than when she looked at it. During examination for referred sensations she reported that when her left knee was touched with a cotton bud, she felt a similar sensation on the plantar aspect of her left foot around the base of her metatarsophalangeal (MTP) joints (Fig. 1b). When this area on her left foot was touched, a similar sensation was experienced in her left knee. The reality of these referred sensations was such that the patient was able to differentiate between light touch and pinprick sensory modalities at both referred sites. There were no referred sensations reported for the non-painful lower limb. Again, when the

### Table 2. Details of all 16 patients

<table>
<thead>
<tr>
<th>Case</th>
<th>Age (yr)</th>
<th>Gender</th>
<th>Disease duration</th>
<th>Affected limb</th>
<th>Pain level on movement at presentation*</th>
</tr>
</thead>
<tbody>
<tr>
<td>1a</td>
<td>28</td>
<td>F</td>
<td>3 weeks</td>
<td>Left hand</td>
<td>8</td>
</tr>
<tr>
<td>2b</td>
<td>34</td>
<td>F</td>
<td>8 weeks</td>
<td>Left ankle</td>
<td>8</td>
</tr>
<tr>
<td>3b</td>
<td>24</td>
<td>M</td>
<td>3 yr</td>
<td>Left knee</td>
<td>8</td>
</tr>
<tr>
<td>4b</td>
<td>41</td>
<td>F</td>
<td>6 yr</td>
<td>Right foot</td>
<td>9</td>
</tr>
<tr>
<td>5b</td>
<td>57</td>
<td>F</td>
<td>4 yr</td>
<td>Left hand</td>
<td>5</td>
</tr>
<tr>
<td>Mean</td>
<td>36.8</td>
<td>4F:1M</td>
<td>2.6 yr</td>
<td>Left ankle</td>
<td>7.6</td>
</tr>
<tr>
<td>6</td>
<td>38</td>
<td>F</td>
<td>6 weeks</td>
<td>Left leg</td>
<td>5</td>
</tr>
<tr>
<td>7</td>
<td>35</td>
<td>F</td>
<td>5 months</td>
<td>Right arm</td>
<td>5</td>
</tr>
<tr>
<td>8</td>
<td>40</td>
<td>F</td>
<td>1 yr</td>
<td>Right arm</td>
<td>6</td>
</tr>
<tr>
<td>9</td>
<td>38</td>
<td>F</td>
<td>3 yr</td>
<td>Left leg</td>
<td>5</td>
</tr>
<tr>
<td>10</td>
<td>27</td>
<td>M</td>
<td>2 yr</td>
<td>Left leg</td>
<td>8</td>
</tr>
<tr>
<td>11</td>
<td>51</td>
<td>F</td>
<td>2 yr</td>
<td>Right arm</td>
<td>7.5</td>
</tr>
<tr>
<td>12</td>
<td>68</td>
<td>F</td>
<td>1 yr</td>
<td>Left arm</td>
<td>5</td>
</tr>
<tr>
<td>13</td>
<td>54</td>
<td>M</td>
<td>4 yr</td>
<td>Left foot</td>
<td>9</td>
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<tr>
<td>14</td>
<td>38</td>
<td>F</td>
<td>7 yr</td>
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<td>10</td>
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<tr>
<td>15</td>
<td>22</td>
<td>F</td>
<td>4 yr</td>
<td>Left foot</td>
<td>9</td>
</tr>
<tr>
<td>16</td>
<td>59</td>
<td>F</td>
<td>10 yr</td>
<td>Left foot</td>
<td>9.5</td>
</tr>
<tr>
<td>Mean</td>
<td>42.7</td>
<td>9F:2M</td>
<td>3.1 yr</td>
<td></td>
<td>8</td>
</tr>
</tbody>
</table>

*Visual analogue 10-cm scale.

bReferred sensations reported.
subject watched the examiner touch her affected limb, no referred sensations were reported. This bidirectional referral of sensations could be evoked at the next two assessments but by week 3, following intensive physio- and hydrotherapy, the subject’s pain and swelling had greatly improved and referred sensations were lost.

Case 3
A 23-yr-old man was referred and admitted for rehabilitation. Three years previously he had woken with a spontaneously swollen left knee. No evidence of arthropathy was found despite full investigation including arthroscopy, synovial biopsy and MRI. He was aware of an extreme burning pain and the knee felt as if a ‘red hot poker’ was touching it. This pain persisted for 8 months and was unaffected by analgesics or steroid therapy. He had a nerve block which improved his symptoms for approximately 15 months. He then noticed colour changes in his left ankle and increasing tenderness. Two weeks later his knee became painful again. He underwent a wide number of different therapies (physiotherapy, TENS, acupuncture, nerve blocks), all of which had little or no effect upon his symptoms.

On admission he complained of intense burning pain in his left knee and ankle to feel ‘heavier’ than his right and his right foot was twice the size of his right, his left ankle slightly enlarged and his toes larger than the rest of his left foot. When he was touched with a cotton bud below his left patella, he complained of feeling the same sensation on the plantar aspect of his left foot in the region of his MTP joints (Fig. 1c). A similar sensation was felt again in his left knee when the same region of his foot was touched. He was unable to differentiate between light touch and pinprick; both evoked the same feeling of ‘discomfort’. The sensations were not present when the subject viewed the examiner touching his limb or when vibration was used.

Throughout this subject’s 3-week in-patient stay, the referred sensations could be elicited. Although his mobility had marginally improved on discharge, his pain continued at the same level and his left knee remained swollen.

Case 4
A 41-yr-old woman was referred to the out-patient department with a 7-yr history of pain in her right foot following a Wilson’s osteotomy. She had had delayed healing post-surgery which had required an extended period of immobilization and, despite tricyclic antidepressant therapy and multiple episodes of physiotherapy, she had experienced persistent pain, primarily around her right MTP joints, ever since. On presentation she described a throbbing pain which extended beyond the site of initial injury and was exacerbated by weight bearing. She had allodynia, hyperalgesia in her right foot and dysesthesia on the lower third of her right shin. There was swelling around her MTP joints and a temperature difference of 0.8°C between her right and left lower legs with her right cooler.

With her eyes closed she perceived her right knee and ankle to feel ‘heavier’ than her left and her right foot to be twice the size of her left. When she was touched with a cotton bud on the sole of her right foot, under her MTP joints, she reported feeling the same sensation in her right calf. When light touch was applied to the anterior of her right knee, in the patellar tendon area, this was referred distally to the dorsum of her right foot (Fig. 1d). The same sensations were evoked with a neurotip but not perceived as sharp in the referral site. Vibration was not referred and all referred sensations were lost when she viewed the area being touched.

Over the next 6 months this woman received a novel treatment of mirror visual feedback [15] and was reviewed monthly. Her pain reduced from 6/10 at rest to 1.7/10 as measured by a visual analogue scale and the perceived excessive swelling of her right foot diminished. The referred sensations found on presentation could not be re-evoked at any of her follow-up appointments.

Case 5
A 57-yr-old woman was referred with a 4-yr history of CRPS affecting her left hand and was admitted for leg with the left cooler. His right leg was completely normal in colour and sensations.

With his eyes closed he perceived that his left knee was twice the size of his right, his left ankle slightly enlarged and his toes larger than the rest of his left foot. When he was touched with a cotton bud below his left patella, he complained of feeling the same sensation on the plantar aspect of his left foot in the region of his MTP joints (Fig. 1c). A similar sensation was felt again in his left knee when the same region of his foot was touched. He was unable to differentiate between light touch and pinprick; both evoked the same feeling of ‘discomfort’. The sensations were not present when the subject viewed the examiner touching his limb or when vibration was used.

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rehabilitation. The condition had occurred spontaneously and persisted despite nerve blocks, physiotherapy, acupuncture and Gabapentin. She had fixed flexion deformities of the fingers on her left hand with an extended index finger and complained of intermittent dystonia. Her hand was swollen with alodynia and hyperalgesia present from her fingertips to elbow. Thermal imaging showed a 1.6°C difference between the right and left forearms with the left cooler.

With her eyes closed she described her left hand, from her fingertips to wrist, as feeling tight and larger than the right. When a cotton bud touched her left upper arm she felt a pulling on her left ear (Fig. 1e). This sensation was felt again when she was touched on the left shoulder but, in addition, she now reported that she felt her left thumb, fingers and wrist were also being touched. The referred sensation in her hand increased her pain at rest, from 5/10 to 8/10 on a verbal 10-point scale and her fingers involuntarily became more clawed. When the cotton bud was moved away from her shoulder the sensations in both her ear and hand disappeared, the pain gradually diminished and her fingers relaxed. The referred sensations in the left thumb, fingers and wrist were re-evoked when the left cheek was touched (Fig. 1f). Pinprick evoked the same sensations as light touch at all of the above-mentioned referral sites, but was not perceived as sharp. Vibration was not referred. When the subject viewed her limbs and face being touched, with the aid of a mirror, she reported a tightening in her left fingers but this was to a lesser extent than when she had her eyes closed. Her hand did not become clawed and her pain levels remained at pre-examination levels. The referred sensation of pulling on her left ear and touch on her left hand were not present with vision.

This woman’s condition remained unchanged throughout her 2-week in-patient stay and the referred sensations remained constant.

Summary
All patients reported referred sensations during examination with their eyes closed. They were experienced in real time and disappeared when stimulation ceased or vision was permitted. When the subjects viewed the area being touched the sensations were either diminished (case 5) or not present and when the symptoms of CRPS resolved (cases 1, 2 and 4), referred sensations were lost. Sensations were referred in a modality-specific manner with touch referred in all cases and pinprick also referred in two (cases 1 and 2). Vibration was never referred. All referred sites were located on body parts immediately adjacent, on Penfield’s homunculus, to the stimulated site.

Discussion
This is the first report of referred sensations in CRPS. The novelty of this finding may be due to clinicians not expecting such anomalous sensations or failing to see the potential significance when patients may have reported them. In addition, examining patients with their eyes closed is not routine clinical practice in rheumatology. Light touch was the main sensation referred and this fits well with reports of referred sensations in other conditions [4, 16]. When these sensations are present in amputees, touch is typically the modality referred, with vibration, pinprick, temperature and stroking sensations less so [1].

Light touch is perceived when Ab fibres (large myelinated) are stimulated, though in CRPS Ab fibre stimulation has been found to elicit the experience of pain [17]. However, vibration is also transmitted by Ab fibres, but referral of this modality was not found in our patient sample. Interestingly, Rommel et al. [11] showed an increase in the touch threshold on the ipsilateral side of the CRPS-affected limb using quantitative sensory testing. They concluded that as this deficit extended beyond the area affected by CRPS it was unlikely that systematic damage was occurring at the primary afferents and this was more likely to be due to changes in processing within the central nervous system. In our study we also found that only those with early CRPS (<8 weeks) felt pinprick referred sensations and this may relate to Rommel et al.’s finding that those with significantly longer disease duration had a higher incidence of generalized sensory deficits [11]. However, it is difficult to state conclusively the significance of this result in the light of the small sample size. Referral of temperature was not assessed in this study.

The locations of the referred sites, in our study population, are consistent with previous reports in other pain conditions [1, 18] and fit particularly well with predicted cortical changes that have been shown to occur within the somatosensory body map [19]. Ramachandran [16] proposed that owing to the location and speed with which referred sensations occur in amputees, such ‘ectopic representations’ following functional remapping were probably due to the unmasking of latent synapses within the cortex, as previously described in primates [20, 21]. These synapses are suppressed when there is simultaneous input from two connected receptors, but with reduced or impaired sensory activation in one area, the connection becomes disinhibited. Recent imaging studies, using magnetoencephalography, in six patients with upper limb CRPS type 1 have also shown changes in the cortical somatosensory map, though it was not reported whether these were associated with referred sensations [22]. There was a significantly shorter distance between the areas representing the thumb and little finger on the somatosensory cortex contralateral to the affected limb than the ipsilateral side. Interestingly, there was no significant correlation between the distance of thumb and finger and the level or duration of pain.

Alternatively, referral of sensations may occur at the spinal level. A large body of evidence shows that sensitization of wide dynamic range neurons at level V of the dorsal horn results in ipsilateral and contralateral enlarged receptive fields, which do not rely on a cortical homunculus [23]. In addition, experimental models of
Peripheral neuropathic pain all demonstrate bilateral spinal cord changes after unilateral nerve damage [24]. However, all of our patients had CRPS type 1, so therefore had no precipitating neural trauma. Their sensations were not referred bilaterally, either from the stimulated site to its contralateral partner (i.e. left hand to right hand) or mirrored on the contralateral side (i.e. from stimulated site to referral site on the unaffected limb). In addition, the speed of referral in terms of disease duration, response time on stimulation and resolution as the condition improved, combined with the magnitude of the sensations, all detract from a purely spinal route. Recent thinking is that CRPS is a disorder that involves both CNS and peripheral nervous system components [25, 26]. This is based on the evidence that some patients respond positively to sympathetic blockade, thereby implicating involvement of the sympathetic nervous system, but conversely, sympathetically maintained pain involves the deep somatic tissue (as demonstrated by our patients report of increased pain on movement) which is the domain of the autonomic system. Therefore isolating one clear route for referred sensations is at present problematical.

The reason for the reduction of sensory input in amputees is clear, but in CRPS, where the affected limbs are hypersensitive, one may expect there to be greater sensory input. One explanation is that in CRPS we are seeing a pathological increase in sensory input from one area and hence encroachment of adjacent brain parts following the relocation of the limb’s representation in the sensory map as suggested by the recent imaging studies [22]. Another proposed theory is that the excessive sensory input from the painful area of the affected limb results in a decreased perception of other sensory input from the remaining half of the body, resulting in a functional ‘neglect syndrome’ as demonstrated by a hemisensory deficit [10]. Conversely, it is possible that the considerable sensory dysfunction within the peripheral parts of the painful limb is registered as a loss and the adjacent areas, on Penfield’s homunculus, now encroach. Whichever scenario occurs, the findings from our five case reports show that the processes underlying referred sensations are reversible over a short period of time. Moreover, these processes do not produce referred sensations in the presence of normal sensory or direct visual feedback. The finding of bidirectional referred sensations is particularly novel (cases 2 and 3) and would be impossible to demonstrate in amputees (where the condition is clearly irreversible).

Visual feedback strongly influenced the experience of RS. It is difficult to elicit whether this was true in patients who described RS following amputation, as the methodology in previous reports is not explicit. Moreover most, but not all, phantom limb sensations (where upper limbs are concerned) involve the face or torso, which are not directly viewable on stimulation. Touch and vision are inextricably linked. Touch is known to influence vision, such as distilling the visual illusion of a three-dimensional object when it is drawn on a flat surface. Equally, in some clinical conditions such as somatosensory loss after stroke, visual feedback of the affected limb during testing can significantly improve reported perception [27]. In addition, recent findings by Taylor-Clark et al. [28] showed that the enhancing effect of vision modulated somatosensory cortical processing. Gregory [29] points out that vision evolved from the simpler processes for touch and that it is possible the somatosensory map is inverted (the feet above the hand) in order to correspond with the inverted visual image on the retina. This ensures that the link between vision and touch is as short as possible. Consequently, when our subjects viewed their limbs being stimulated it would appear that the more powerful sense of vision overruled the referred sensations.

The incidence of referred sensations in CRPS was previously unknown, but in this cross-sectional study they were shown to be present in approximately a third of the total study population. Further work, on larger populations, is now required to try to identify any factors that may contribute to the existence of referred sensations and whether their presence is significant to the course of the individual’s disease.

In conclusion, the existence of referred sensations in patients with CRPS type 1 provides evidence of associated central sensory plasticity resulting in or from impairment to peripheral neural systems.

**Conflict of interest**

The authors have declared no conflicts of interest.

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