Findings that shed new light on the possible pathogenesis of a disease
or an adverse effect

Somatotopic mismatch following stroke: a pathophysiological condition escaping detection

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Summary

Clinical evaluation of somatosensory deficits in stroke patients is very limited and usually does not include testing of somatotopic organisation, which is a prerequisite for meaningful interpretation of sensory input and sensorimotor control. Detailed tactile testing of the left hand of a 54-year-old patient suffering from sensory deficit and central pain after a right-sided stroke revealed severe distortion of somatotopic sensory maps as evidenced by incorrect localisation of the point stimuli. Unlike previously reported gross somatotopic remapping taking place within reduced representational space after lesion, this is the first case report revealing chaotic scrambled somatosensory maps. While the incidence of such scrambled somatotopic representation of tactile input is not yet known in stroke patients, current observations indicate that in-depth investigations of somatotopic organisation of affected area may reveal the underlying cause for various functional deficits including central pain. Thus, new rehabilitation strategies may need to be developed specifically for such patients.

BACKGROUND

Rehabilitation strategies aimed at regaining dexterity after stroke largely focus on motor recovery; however, dexterity of human hand essentially relies on intact sensory input provided by tactile mechanoreceptors. Without tactile sensory information functional use of the hand is hardly possible.^{1–3} To be able to extract information from spatio-temporal pattern of afferent responses signalling properties of objects touched or held in a hand, somatotopic representation of hand's skin has to be preserved.

We report here a patient suffering from somatosensory and sensorimotor impairments of her left hand following right middle cerebral artery territory infarction. Two of the most severe are tactile agnosia, that is, necessity to engage vision for control of hand function, and tactile allodynia. The complex of symptoms still present 10 years after the incident indicates that underlying cause of functional impairments is likely to be due to sensory dysfunction. This prompted us to perform detailed tactile testing which revealed loss of organised somatotopic representation of hand as evident from the referred sensations and incorrect localisation of point stimuli on the affected hand.

CASE PRESENTATION

A previously healthy 45-year-old woman was hospitalised in 2002 with the infarction of the right middle cerebral artery territory. Stroke was caused secondary cerebral vasospasm which followed aneurysmal subarachnoid haemorrhage. MRI scan of the head demonstrated right middle cerebral artery infarction mainly affecting parietal and temporal lobe as well as internal capsule (figure 1).

After 10 years, fine hand actions are still impossible without visual endorsement and visual movement tracking. In everyday life, the patient has enough motor strength to use the affected hand to hold objects or even a shopping bag; however, when out of sight, the hand may spontaneously release the grip completely without patient being aware of that. All those observations indicate that underlying cause of functional deficits is likely to be sensory rather than motor.

Central pain in terms of tactile allodynia is another leading symptom. Interestingly, evoked pain is less severe when patient has expected or sees the application of the stimulus, but more severe when not prepared for the stimulus or when touched by haptically complex or unfamiliar objects. Intensity of pain does not correlate with intensity of stimulus: the patient tolerates well strong pressure such as applied during massage while gentle touch or water drops may cause severe pain. This means that allodynia in this patient is caused by the activation pattern of tactile afferents rather than the response intensity of specific afferent type.

INVESTIGATIONS

Measurement of detection thresholds

Mechanical detection thresholds were measured with Semmes-Weinstein Monofilaments (Touch-Test Sensory Evaluators; Stoelthing Co., Wood Dale, Illinois, USA), using a series of increasing/decreasing stimuli. The patient could not detect the filament calibrated for 0.16 g force (evaluator size 3.22) at the fingertips of her left hand. Filaments exerting 0.6 g force (evaluator size 3.84) were

Figure 1 Structural MRI scan. MRI scans of the head showing right middle cerebral artery infarction mainly affecting parietal and temporal lobe. Black artefacts are induced by metal residual after operation and aneurysm clip.

identified in about 75% of all attempts; however, the patient remained unsure in her responses until reaching filament calibrated for 1.4 g force (evaluator size 4.17) at which patient felt confidence in her report.

On the contralateral healthy hand, mechanical detection threshold was 0.02 g (evaluator size 2.36) which is within the normal range.⁴ No obvious differences between five fingers of either hand were seen.

Testing for somatotopic organisation

In this test the patient had to indicate the location where the touch by the monofilament was felt. Tests were performed with filament calibrated for 4 g, which was determined to be above the detection threshold, and was clearly felt in all parts of the hand. Twenty-five predefined different test sites on the glabrous skin of the hand were tested in numbered order by the examiner according to the protocol as shown in figure 2A. Data for each test site were obtained only once as it required a lot of concentration from the patient. During stimulation the patient had her eyes closed. If stimulus was felt, the patient opened eyes and pointed to the location on the skin where

stimulus was felt. The subject also had to report on the size of the object's contact area, whether the border of contact area was sharp or diffuse and whether the patient was convinced about the location where touch was made. To avoid false reports by unprovoked spontaneous sensations, the time of stimulation since the patient closed her eyes was randomised and the patient was asked to say 'yes' immediately when stimulus was felt. If no sensation was reported then filament was reapplied to exactly the same spot after random interstimulus interval (>1 s). If no sensation was reported after the third application, it was regarded as no stimulus being felt. There was always a good temporal match between actual stimulation and the patient's indication of sensation.

Of 25 test sites of the left hand, the patient did not feel any stimulation at only one site (figure 2B, #22) while stimuli at 4 other sites were clearly felt but could not be localised with confidence (#6, #15, #18 and #21) and therefore were not analysed. Three of those stimulation points were on the ring finger; however, evaluation of detection thresholds on this finger did not show any difference in comparison with other fingers. For the majority

Figure 2 Test for the ability to localise tactile stimuli. (A) Twenty-five test sites on glabrous skin of the left hand. (B) Stimulus localisation mismatch. Arrows originate from the centre of each test site where stimulus was applied and terminate at the location where stimulus was felt. At test site #4 stimulus was felt exactly at the location where it was applied (encircled). Stimuli at test sites #6, #15, #18 and #21 were felt, but could not be localised with confidence. Stimulus at test site #22 (crossed) was not felt at all.

of stimuli 20/25 (80%), the patient was confident of the location where the skin was touched; however, the patient made systematic errors when asked to point to the stimulation site on the skin. Those observations indicate that the patient experienced referred sensations. Importantly, out of 13 stimuli, which were applied to the fingers, none was felt at the same phalanx where the stimulus was applied. Out of seven stimuli applied to the palm, only one (#4) was localised correctly, four other points were mislocalised 3–14 cm away from the actual stimulation site without any systematic pattern.

Contact area of 8 stimuli whose location could be identified was felt as a small well-defined dot while the remaining 12 stimuli were felt as an area about 1 cm in diameter with fuzzy borders. For all detectable stimuli the quality of the stimulation was described as non-painful touch.

In a separate control experiment the repeated stimulation of three stimulation sites demonstrated that there was fair consistency in mislocalisation of the same stimulation site locations thus supporting conclusions based on patient's report that those were true referred sensations and not just guessing of the location of stimulus. The three chosen stimulation sites were felt as a small well-defined dot and were tested intermingled with stimulation at the other random locations.

On the contralateral healthy hand, the patient could indicate the exact location of all 25 stimulation points.

DISCUSSION

In our patient, pathological central reorganisation following stroke has resulted in loss of somatotopic organisation. Unlike similar reports showing expansion/shrinkage or systematic shift in somatotopic representation,^{5–8} our study shows scrambled disordered somatotopic maps at

local or microscale without any systematic pattern.⁹ We believe that this is the reason why this scrambled somatotopic representation remained undetected by all routine neurological examinations and remained unknown to the patient herself. Given the somatotopic organisation of ventroposterior nucleus of the thalamus¹⁰ and widespread connections with somatosensory cortex,¹¹ it is likely that a scrambled somatotopic representation might be fairly common disorder, but it has not been specifically searched for or easily noticed. Study by Bender *et al*¹² comprising data from 558 patients has shown that one of the most sensitive tests to detect sensory disturbances due to a wide range of underlying causes are tests for graphesthesia and directional cutaneous kinesthesia. Both tests due to their critical dependence on orientation of cutaneous sensory space would readily detect abnormalities caused by scrambled somatotopic maps; nevertheless, they would not identify the underlying mechanism behind those functional deficits. Simple testing procedure demonstrated in our case study shows the way in which to differentiate patients with disturbances in somatotopic organisation. This is a crucial step to choose the most efficient rehabilitation strategy and understand associated complex sensorimotor deficits as spatiotemporal pattern of afferent activation is crucial for normal tactile sensory function and motor control.^{1–13} For example, study by Blennerhassett *et al*³ on 45 stroke patients has convincingly demonstrated how deficits in ability to sense particular object properties like friction are manifested in the grip force control dysfunction which is not related to poststroke motor deficits *per se*.

We suggest that scrambled somatosensory maps may preclude the brain from making meaningful interpretation of the sensory input and thus is the underlying cause of impaired sensorimotor control of hand as clearly

evident in our patient. Furthermore, abnormal and uninterpretable sensory input may also cause non-noxious stimuli to be perceived as painful and might therefore cause central pain in our patient.¹⁴ In this and similar patients, methods targeting normalisation of somatotopic representation by means of learning or multisensory integration may prove to be the most effective approach to functional rehabilitation and treatment of neuropathic pain. For example, this might explain why mirror-therapy can be effective in treating somatic pain syndromes and in some patients is beneficial to achieve sensorimotor recovery after stroke.¹⁵ Nevertheless, it is not known as to whether there is a critical time window for intervention to be successful. It also remains to be seen whether normalisation of somatotopic representation can be achieved as long as 10 years after the stroke and whether early detection would improve chances of recovery. Raised awareness of this condition may help to identify more such patients and answer those questions in systematic studies.

Learning points

- Somatosensory dysfunction could be one of the key factors determining poor recovery of hand dexterity after stroke.
- Disordered scrambled somatotopic representation may preclude meaningful interpretation of tactile sensory input. While consequences of such deficits are readily detected, the routine neurological examinations are unlikely to identify the underlying cause and reveal somatotopic mismatch.
- In this and similar patients rehabilitation strategies promoting normalisation of somatotopic representation are expected to improve hand dexterity and probably treat allodynia.

Competing interests None.

Patient consent Obtained.

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