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REVIEW ARTICLE

Paradoxes in rehabilitation

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ABSTRACT
Paradoxical enhancement and paradoxical recovery of function after brain injury harmonize well with the concept of "ultrabilitation" and its focus on novel forms of flourishing in rehabilitation settings. I consider three sets of paradoxes which may impact on brain injury rehabilitation. Firstly, I consider post-traumatic growth after brain injury and its key determinants. Secondly, I review the role of illusions in rehabilitation and the paradox that some clinical conditions may be improved by invoking perceptual distortions. Thirdly, I consider paradoxical recovery profiles after brain injury, since knowledge of such paradoxical profiles may help inform attempts at rehabilitation of some patients. Finally, I consider how some of these paradoxes relate to components of ultrabilitation, and in addition to the nascent field of positive neuropsychology and the concept of resilience after brain injury.

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➤ IMPLICATIONS FOR REHABILITATION

- Illusions can sometimes be harnessed as a therapeutic tool in rehabilitation.
- There may be spontaneous, positive outcomes of an injury or illness, in the form of "post-traumatic growth", and these should be considered as part of a holistic therapeutic approach in rehabilitation.
- Some patients make an exceptional recovery from a severe brain insult, and lessons could be learned from such cases, such as disciplined use of compensatory strategies, which could have broader implications for neurorehabilitation.

The way of paradoxes is the way of truth – Oscar Wilde, 1891

Introduction

"Ultrabilitation" is a new and promising concept which promotes the notion of flourishing in rehabilitation settings, with a focus on moving beyond or around recovery and permitting novel forms of adjustment which complement the more standard outcomes relating to alleviation of functional deficits. Paradoxical enhancement and paradoxical recovery of function after brain injury fit in well with the concept of ultrabilitation, and both sets of concepts form the perfect catalyst for encouraging creative forms of adjustment after brain injury.

Paradoxes have played a role in many revolutionary discoveries, sometimes leading to Nobel Prizes. For example, Chandrasekhar's Physics Nobel Prize Lecture [1] referred to a resolution of the "Eddington Paradox", named after the famous English astrophysicist, whereby a star which had cooled to absolute zero somehow found the energy to undergo major expansion. Some time later, in 2004, the Nobel Prize for Physics was won by Frank Wilczek. His prize lecture was entitled *Asymptotic Freedom: from Paradox to Paradigm* [2]. Wilczek referred to paradoxical findings in physics that gave rise to the discovery of a new dynamical principle, "asymptotic freedom". Paradoxical phenomena abound in nature [3], and are evident in fields of inquiry such as epidemiology [4,5], human aging [6], child development [7], cognitive psychology [8], and psychological well-being [9].

"*Neurology's favorite word is deficit, denoting an impairment or incapacity of neurological function*", noted Oliver Sacks [10, p.1]. Ten years later, in *An Anthropologist on Mars*, he wrote, "*Defects, disorders, diseases, in this sense, can play a paradoxical role, by bringing out latent powers, developments, evolutions, forms of life, that might never be seen, or even be imaginable, in their absence*" [11, p.xii]. I do not refute the fact that, for most people, neurological impairment reduces their possibilities within the world, and frequently requires them to re-learn functions and actions that previously occurred automatically. However, I suggest that these losses are not always the only outcomes of nervous system insults. The aim of this article is to explore examples of paradoxes which may have implications for rehabilitation, and which have resonance with the concept of "ultrabilitation" [12]. I consider three topics – post-traumatic growth, the role of illusions in neurorehabilitation and paradoxical recovery profiles in neurological conditions. I conclude with a more general consideration of how such paradoxes may inform the nascent field of positive neuropsychology, and may inform the role of resilience in recovery from a brain insult.

Post-Traumatic growth

The spiritual eyesight improves as the physical eyesight declines – Plato
Sweet are the uses of adversity – Shakespeare
What does not kill me makes me stronger – Nietzsche

In recent years there has been an increased focus on the notion that positive changes may be seen in some patients who survive

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a brain injury or brain illness – "post-traumatic growth" (PTG), with an "explosion" of books on this topic (e.g. [13–16]). There are a number of definitions of post-traumatic growth, and the pragmatic one that I will use is – *Unanticipated positive adjustment following a single adverse event or a period of adversity*. Post-traumatic growth represents a form of flourishing after a significant adverse event, going beyond the standard recovery process. It is a subset of what can be encompassed under the concept of "ultrabilitation", since the latter would also include non-traumatic conditions.

Post-traumatic growth may include changes for the better such as greater appreciation of life and nature, warmer relationships with others, an improved sense of priorities, greater achievements in work or personal settings and enhanced self-confidence to deal with adverse events in the future. Scales have been developed to assess PTG, such as the Post Traumatic Growth Inventory [17]. Some authors have attempted to apply the concept more generally to psychiatric disorders and to adverse life events [18,19], and to conditions such as cardiac disease [20]. While the concept of PTG also applies to disciplines other than neurology [21], with similarities noted in PTG for those with acquired brain injury and those with myocardial infarction [22], relevant observations in neurological conditions are particularly paradoxical in view of the generally negative outcomes that are highlighted after an insult to the brain. In the case of mental stress, Taku et al. [23] attempted a factor analysis of the key components of post-traumatic growth, and identified five factors – Relating to Others, New Possibilities, Personal Strength, Spiritual Change, and Appreciation of Life. It is of course important to see such instances of "post-traumatic growth" in the context of the usually disabling and distressing nature of adverse life events such as psychiatric and neurological disorders, and to take account of general reservations such as those outlined by Coyne et al. [24] in the case of cancer patients, but these instances of positive outcomes in clinical disorders may offer clues as to what may or not be fruitful avenues in therapeutic intervention.

Both McGrath and Linley [25] and Powell et al. [26] noted that in a group of patients with TBI, long-term follow-up indicated that post-traumatic growth appeared to increase over time after the head injury. In a subsequent follow-up study, Powell et al. [26] found no changes in PTG between 11 and 13 years post-injury, and this suggested that once PTG is established it is a relatively stable phenomenon. Hawley and Joseph [27] found that on long-term follow-up (an average of 11 years) after traumatic brain injury, around half of their participants showed evidence of post-traumatic growth on a structured questionnaire, responding positively to items such as – "*I don't take life for granted any more*" and *"I value my relationships much more now*". Hawley and Joseph [27] did not find significant correlations between PTG and injury characteristics such as severity of TBI. Sekely and Zakzanis [28] examined a group of mild TBI patients and found that around a third of the sample exhibited PTG, with the most common PTG features being a greater appreciation of life, better relationships with others, and enhanced personal strength. Karagiorgou et al. [29] looked at PTG in a sample of TBI patients undergoing a positive psychotherapy intervention, and noted that this group showed two components of PTG, lifestyle improvements and exploring new possibilities. In a meta-analysis of studies looking at PTG in TBI, Grace et al. [30] found that PTG was related to being in employment, having a longer period of education, positive cognitive appraisal of the injury, being in a relationship, older age, longer time since injury and lower levels of depression.

As well as in TBI, PTG has also been systematically studied in stroke survivors [31–33] and also in stroke carers [34]. In stroke survivors, PTG has been associated with a high level of reflection, social support and university education [35]. Denial as a coping strategy has been associated with PTG in stroke [31], and the authors also noted links with positive cognitive restructuring, downward comparison and resolution. Downward comparison, whereby as a cognitive coping strategy individuals reflect on others who are less well off, has been explored more generally in studies of mental well-being (e.g. [36]). Resolution refers to accepting and coming to terms with adverse events. Denial as a coping strategy has also been noted in stroke carers [34]. Kelly et al. [37] found that PTG was evident four to six months after stroke, and that key factors in the development of PTG included deliberate rumination, active coping and denial coping.

The value of illusions in neurorehabilitation

The brain constructs and regularly updates its own models of the world which allow fast and accurate perception, efficient memory, successful decision-making and appropriate action. If those models are wrong in significant ways, the consequences can be very serious. Accordingly, perceptual illusions such as visual and auditory hallucinations, distorted spatial representations of the world and cognitive illusions normally have a highly disruptive effect on thinking and behavior. Here, however, I consider some ways in which deliberately inducing illusions, with the aid of simple technologies, may paradoxically improve function in patients with some form of disability. The use of technologies to go beyond usual sensory boundaries is considered to be an important set of conditions for "ultrabilitation" to occur [12].

In this section, I consider two illusions, the mirror box illusion and the rubber hand illusion. For a more general discussion of illusions in neurorehabilitation, the reader is referred to the chapter by Manly et al. [38], and for a recent review on the use of prism adaptation on visual neglect, a topic covered by Manly et al, the reader is referred to a recent systematic review by Champod et al. [39].

Mirror Box Illusion

One common illusion to which we are all prone is the idea that, when our foot hurts, the pain we experience is in our foot rather than in our brain. This illusion is no more dramatically revealed than when, following limb amputation, people experience crippling pain in the now absent limb. This "phantom limb pain" is disturbing, not least because normal strategies such as rubbing or flexing the hurt area are not available. A novel approach to helping such patients has been to create the illusion that the absent limb is still present. This is achieved using a mirror to reflect the remaining limb in the location of the missing limb, often using a mirror box, hence the term Mirror Box Illusion. Remarkably, despite the patients knowing that they are not seeing their absent limb, over a large number of trials the illusion of cramp relieving movement can reduce the pain, sometimes permanently [40–41]. A randomized control trial [42] found that mirror therapy was effective in reducing the intensity, duration and frequency of phantom limb pain. Presumably, the "latent" cortical map of the missing limb somehow gets activated. In some amputees, however, phantom limb movement can actually result in an increase in phantom limb pain, and a variation of the Mirror Box Illusion has been developed, whereby patients look at the mirror reflection of touches applied to the intact hand, while at the same time receiving touches on the remnant stump which is positioned behind the mirror. The illusion that is generated – of touch sensations on the phantom hand – has been found to reduce phantom limb pain in patients who do not respond to the standard mirror box illusion [43]. It may therefore be the case that different forms of the illusion will help different patients.

Ortiz-Catalan et al. [44] found relief of phantom limb pain and less intrusion into daily activities, including sleep, as a result of an intervention which included motor execution of the phantom limb that was aided by machine learning and augmented virtual reality. A recent single-blind randomized control trial [45] sounded a cautionary note with regards to the use of mirror therapy for phantom limb pain, with the authors finding that, although there were some individual differences in patient subgroups, mirror therapy was little better than sensory-motor exercises of the intact limb without a mirror, and having an augmented reality component to the mirror therapy did not yield any specific benefits. In addition, a recent systematic review [46] pointed to the relative absence of high quality evidence for the effectiveness of mirror therapy and related feedback interventions for the relief of phantom limb pain. However, the evidence for the value of such paradoxical interventions remains strong, even though they may not be appropriate in all settings.

In addition to alleviating phantom limb pain, the Mirror Box Illusion has also been used to treat Complex Regional Pain Syndrome, pain which usually lasts for at least six months and affects one limb. As well as excessive pain, there may be changes in skin color, temperature or swelling in the affected area. In a randomized control trial, Vural et al. [47] found significant improvements in stroke patients with this pain syndrome when the Mirror Box Illusion was combined with standard therapy, compared to those who just received standard therapy.

Apart from the relief of pain, the other main application of the Mirror Box Illusion or more generally mirror visual feedback has been in the rehabilitation of hemiparesis in stroke patients [48]. It would appear that watching the reflection of self-generated movements in a mirror results in improved self-awareness and spatial attention, and this may directly or indirectly stimulate recovery. Arya et al. [49] found that 40 sessions over an eight week period produced significant gains across several upper limb motor recovery measures in the mirror box therapy group compared to the control group, and in a later study [50] found similar benefits for lower limb recovery and gait. In a systematic review of the use of mirror therapy to aid recovery in stroke, Yang et al. [51] found evidence to support the beneficial effects of mirror therapy on motor function, activities of daily living and relief of pain. Finally, in one study of unilateral visual neglect, therapy that included the Mirror Box Illusion over a four week treatment period resulted in improvement in measures of neglect [52].

Rubber hand illusion

In the rubber hand illusion, a person's hand, hidden from view, is brushed by a third party and at the same time a rubber hand that is fully visible is also brushed. This results in the individual feeling a sense of ownership of the rubber hand, and sensations from the brushed rubber hand are projected to the individual. Although there is only limited evidence on the value of the rubber hand illusion in neurorehabilitation [53], the illusion of body ownership finds a natural application in those patients who suffer from body ownership disorders, such as somatoparaphrenia, where the patient may deny ownership of a limb or the entire side of his/her body, with such patients showing improvement in their condition when exposed to the illusion [54,55]. The rubber hand illusion encourages tactile awareness, perhaps by reactivating tactile memories, and it has found application in the complex regional pain syndrome referred to above [56], and in cervical spinal cord injury [57]. One study [58] found short-term improvement in unilateral visual neglect when the patient experienced the rubber hand illusion, presumably by cueing spatial attention or changing egocentric reference frames.

Paradoxical recovery profiles

Post-traumatic growth, covered above, could be classed as a paradoxical recovery profile, since the patient flourishes and shows enhanced well-being or adjustment in the context of a clinical condition where deficits and negative consequences generally abound. There are also paradoxical recovery profiles such the Double-Hit Recovery phenomenon, whereby a second lesion can abolish or mitigate the deleterious effects of the first lesion [59,60], and the Delayed Deterioration paradox whereby there appears to be good initial recovery following a brain insult and then an unexpected dramatic decline in function [61–63]. For reasons of space, I will limit this section to two paradoxical recovery profiles – late recovery from disorders of consciousness, and exceptional recovery after brain injury, both of which contrast with the normal trajectory of recovery, namely for those with a severe insult to the brain to show major deficits early in the recovery period, and thereafter a gradual recovery followed by a plateau, the end point of which is characterized by significant residual impairments.

Unusually late recovery in disorders of consciousness

While most brain injury patients with prolonged periods of severe disorder of consciousness, such as prolonged vegetative state, will have a poor outcome (e.g. [64]), both group and single-case studies have reported cases of late recovery from an initial state which appeared to point to very poor prognosis. Estraneo et al. [65] looked at long-term outcome (around two years after brain injury) in 50 patients with an initial vegetative state for at least six months. They found late recovery in 25% of patients, and in this case "late" was generally two years or more post injury. In a later study from the same group [66], long-term outcome (5–6 years post-injury) was examined in 13 of those cases who recovered from a vegetative state one year after a severe TBI or six months after a non-traumatic brain injury. Of these 13 cases, two had died, five were still in a minimally-conscious state and six recovered full consciousness. Although these six patients had marked cognitive impairments, most were able to meaningfully interact with family members and care staff. Yelden et al. [67] followed up 34 patients 2–16 years after they were diagnosed with prolonged disorders of consciousness (seven in a minimally conscious state, 27 in a persistent vegetative state). Of these 34 patients, 32% showed late improvement of awareness, albeit remaining severely disabled. Most of these late recoveries (45%) occurred in patients with subarachnoid hemorrhage. In a single-case study, Wilson et al. [68] reported a case of a patient with TB meningitis who was mute, stuporous and barely more than minimally conscious for more than two years after her brain insult. Around the time of cessation of her TB medication (which may of course have been a coincidence), her level of consciousness improved to the point where she could cooperate with neuropsychological testing, and after further rehabilitation she could walk with a zimmer frame, was largely continent and could eat with minimal assistance and prompting. A further example of delayed recovery was described in a book devoted to the case [69] of a patient who suffered a severe brain injury at the age of 28 years following an assault. He was in a state of disordered consciousness for 19 months, with a very poor prognosis. However, with intensive rehabilitation input he then made a good recovery, with functions such as speech and mobility showing major improvements, and he even had above-average performance on the WAIS Block Design subtest.

Exceptional recovery after a severe brain injury

While the vast majority of studies of brain injury, whether it be traumatic or non-traumatic, focus on the imperfect recovery of patients, characterized by residual deficits and difficulties, it is important to see if we can learn any lessons from those cases of apparently exceptional recovery after a severe brain injury. Schutz [70] documented a series of nine such cases, and suggested that it was the pursuit by these patients of compensatory strategies with "uncommon persistence and self-discipline" which may have been a key factor in their excellent recovery. Schutz and Schutz [71] reached similar conclusions in their analysis of profiles of recovery in a larger series of TBI cases. A single-case study found what was considered to be exceptional recovery [72] in a 13-year old football player who had a primarily compressive lesion (acute subdural haematoma), and it is possible that both the young age and the compressive nature of the lesion may have been key factors in recovery.

An interesting group of patients who are often seen to fall into the category of exceptional recovery are doctors or other senior professionals who suffer a severe brain injury, although one might argue that they write about their recovery more than other TBI victims. Some of these cases, encompassing a range of etiologies were documented by Kapur [73] in his book, *Injured Brains of Medical Minds: Views from Within*. Specific additional case studies have been reported since the publication of that book (e.g. [74–80]). Together, these articles appear to have a common theme of positivity and perseverance. These accounts naturally raise the issue as to how important is "cognitive reserve" in influencing the recovery process. Although this is a topic in its own right, suffice to say that the range of evidence (e.g. [81–84]) appears to support some role for level of premorbid cognitive functioning in determining recovery, if only because the factors outlined above may be more likely to come into play, factors such as extensive use of compensatory strategies and aids (which may result in creative solutions that lead to enhanced performance), having a positive outlook and showing self-discipline and perseverance in the course of the rehabilitation process.

Conclusions, positive neuropsychology and resilience

The darker the night, the brighter the stars – Fyodor Dostoyevsky

In this final section, I provide an overview and a framework which may help the concepts discussed above to sit more broadly within the field of neurorehabilitation. "Ultrabilitation", with its focus on flourishing and enhanced adjustment in the recovery process, fits comfortably with the domains of positive neuropsychology and resilience, and I therefore cover these topics in this section.

Influential views within contemporary neuroscience view the brain as a dynamic, adaptive and evolving system shaped by its environment and itself. Damage to the brain may upset one dynamic state and lead to unpredictable effects, some beneficial and some harmful, as it settles into a new state [85]. An interesting development in recent years has been a focus on one potentially beneficial side-effect of a brain insult, namely hyperconnectivity. This is broadly defined as enhanced functional connectivity between brain networks. Hillary and Grafman [86] have reviewed some of the neural mechanisms underlying such changes in connectivity, which may be linked to both impaired and enhanced cognitive performance. From the rehabilitation perspective, it is worth noting evidence linking what appear to be compensatory structural brain changes to creativity in dementia [87], and hyperconnectivity features which have been implicated in enhanced performance in some individuals with autism [88] and in musicians with absolute pitch [89]. It is possible to speculate that some cases of paradoxical recovery may be related to compensatory neural changes such as hyperconnectivity.

Without minimizing the very real losses that often arise after brain injury, the paradoxical phenomena noted in this article emphasize the importance of looking at overall change rather than simply focusing on deficit. The "disability paradox", whereby those with disability report higher levels of happiness than healthy individuals asked to estimate the happiness of the disabled individuals [90,91], is one example where preconceived negative biases can lead to states of wellbeing being ignored or undervalued. This echoes a broader approach of positive psychology and positive clinical psychology that places emphasis on dispositional optimism, flourishing, and functional reserve in coping with impairment [92,93]. These fields suggest that we focus on intact skills, on past strengths and interests, and on how both rehabilitation efforts and residential environments (cf. [94]) can be altered to take these skills and talents into account. I would like to propose that the paradoxes noted in this article can help to point to a paradigm shift in how we view illnesses of the brain, and, in particular, point to a new, emerging field, that of positive neuropsychology, and more generally to the fields of positive neurology [95] and positive neuroscience (see Greene at al., [96] and the website www.posneuroscience.org). Randolph [97] has identified six domains where positive neuropsychology may have an impact on wellbeing – use of compensatory strategies, activity engagement, prevention of cognitive problems, public education, understanding positive outcomes in neuropsychiatric populations and studying individuals with robust cognition. Rabinowitz and Arnett [98] have reviewed evidence that shows how TBI recovery can be enhanced by a focus on positive psychological phenomena such as positive affect, optimism, grit and adaptive coping style, and similar promising findings have been reported by Cullen et al [99].

A final positive neuropsychology element which stems from some of the paradoxes reviewed in this paper relates to resilience after brain injury, and one could argue that this is necessary component of the flourishing side of "ultrabilitation". Resilience is generally taken to refer to processes, usually psychological but which could also be biological, by which individuals reduce the potential negative impact of adversity, ranging from a single adverse event to life-long adversity related to a developmental condition. The importance of resilience in TBI is highlighted by its close relationship to positive outcome such as greater community participation [100]. In a longitudinal study of resilience after TBI, Marwitz et al. [101] found that higher levels of resilience were related to nonminority status, absence of pre-injury substance abuse, lower levels of anxiety and disability, and greater life satisfaction. Neils-Strunjas et al. [102] have reviewed four tools to assess resilience after brain injury, and have provided a framework comprising five domains of resilience – personal strengths, strengthening experiences, supportive relationships, a sense of control and extracting meaning from life experiences. Nalder et al. [103] have proposed a resiliency model to help frame targets and desired outcomes in neurorehabilitation. The six components to this model are – TBI-related adversity; initial response to the adversity; personal affective, cognitive and behavioral characteristics which may serve as protective factors; resources that represent environmental protective factors; affective, cognitive and behavioral self-regulatory processes; enabling beliefs that promote positive thinking; and resiliency-related outcomes. Hanks et al. [104] examined factors contributing to resilience in a group of TBI patients ranging in severity of injury. While they did not find that resilience was determined by premorbid intelligence, brain injury severity or cognitive flexibility, they did find a relationship with coping style as assessed by a coping inventory, noting that task-oriented, problem-solving coping strategies were more closely related to resilience than avoidance-coping strategies. In addition, they found resilience related to perceived social support – how individuals felt that social networks provided resources consistent with their needs and expectations. Finally, resilience may be related to practice in exercising cognitive control mechanisms, as in inhibiting particular memories [105,106].

In conclusion, paradoxical phenomena in brain injury challenge our thinking about neurological conditions and the after-effects of a brain insult. They suggest new therapeutic avenues and harmonize with the concept of ultrabilitation [12]. I hope that researchers and clinicians alike will respond to the challenge and gather empirical data to help inform and progress treatment interventions which derive from such paradoxes, and at the same time advance the concept of ultrabilitation.

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