Neuroscience and Biobehavioral Reviews 80 (2017) 729–742

Contents lists available at ScienceDirect

Neuroscience and Biobehavioral Reviews

journal homepage: www.elsevier.com/locate/neubiorev

Review article

Conceptualising compensation in neurodevelopmental disorders: Reflections from autism spectrum disorder

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ARTICLE INFO

Keywords:
Autism Spectrum Disorder
Neurodevelopmental disorders
Compensation
Compensatory mechanisms
Adaptation
Camouflaging
Cognitive phenotype
Behavioural phenotype
Remediation
Theory of mind
Executive function
Good outcome
Under-diagnosis
Late diagnosis
Female presentation
Unaffected siblings

ABSTRACT

Within research into neurodevelopmental disorders, little is known about the mechanisms underpinning changes in symptom severity across development. When the behavioural presentation of a condition improves/symptoms lessen, this may be because core underlying atypicalities in cognition/neural function have ameliorated. An alternative possibility is 'compensation'; that the behavioural presentation *appears* improved, despite persisting deficits at cognitive and/or neurobiological levels. There is, however, currently no agreed technical definition of compensation or its behavioural, cognitive and neural characteristics. Furthermore, its workings in neurodevelopmental disorders have not been studied directly. Here, we review current evidence for compensation in neurodevelopmental disorders, using Autism Spectrum Disorder as an example, in order to move towards a better conceptualisation of the construct. We propose a transdiagnostic framework, where compensation represents the processes responsible for an observed mismatch between behaviour and underlying cognition in a neurodevelopmental disorder, at any point in development. Further, we explore potential cognitive and neural mechanisms driving compensation and discuss the broader relevance of the concept within research and clinical settings.

1. Introduction

Much research into neurodevelopmental disorders has focused on uncovering the cognitive and neurobiologial atypicalities that underlie the defining behavioural symptoms of the condition in question. In the case of Autism Spectrum Disorder (ASD), numerous cognitive theories (e.g., deficient 'theory of mind', 'weak central coherence'; see Frith, 2012) have been proposed to account for the social and non-social symptoms that are characteristic of the condition. And yet, across neurodevelopmental disorders and pertinent in the case of ASD, there is great heterogeneity in the degree to which symptoms lessen, persist or even worsen across the lifetime. The mechanisms by which the behavioural presentation of a condition might alter across development remain largely elusive. For example, in the case of ASD, there currently exists no empirically-grounded explanation pertaining to why some autistic children no longer fulfil diagnostic criteria by adulthood and likewise, why some autistic individuals do not present with clinically impairing symptoms *until* adulthood.

It is tempting to assume that any significant improvement in the behavioural presentation of a neurodevelopmental condition has come about due to some alteration or alleviation of atypical underlying cognition and/or neural function. In an authoritative opinion piece on the last 25 years of research into ASD and developmental dyslexia, Uta Frith highlighted that "it is still not clear what causes these changes and wide variations [in behavioural symptoms]…but compensation makes it possible to disguise persisting problems" (2013, p. 670). This begs the question; what is compensation and how can we measure it? Within the field of neurodevelopmental disorders, the phenomenon remains relatively abstract and ill-defined, such that numerous, potentially overlapping terminologies have been used in the literature (e.g., in ASD, camouflaging/masking, Lai et al., 2016; compensatory learning, Frith, 1991; adaptation, Johnson et al., 2015b), but the construct has never been directly studied in its own right. Nevertheless, the importance of understanding compensation in neurodevelopmental phenotypes is clear. First, given increasing interest in studying previously conceived 'childhood' disorders, such as ASD and Attention Deficit Hyperactivity Disorder (ADHD), across the whole lifespan, including how disorder presentation might change over time (for ASD, see Georgiades et al.,

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http://dx.doi.org/10.1016/j.neubiorev.2017.06.005
Received 1 March 2017; Received in revised form 25 May 2017; Accepted 13 June 2017
Available online 19 June 2017

2017), developmentally-relevant mechanisms for improvement, including compensation, should be an important focus of research. Second, compensation could be a useful way to unpick some of the heterogeneity amongst neurodevelopmental disorders, which is frequently proposed to be one of the greatest challenges to understanding these conditions (Thapar et al., 2017). Finally, studying the mechanisms underlying compensation could be fundamental to informing early intervention research, whose principle aim is to improve long-term prognosis. And yet, in order to directly investigate compensation, we must have a reasonable definition to guide our measurements and derive testable hypotheses.

In this paper, we aim to create a working definition of compensation relevant to neurodevelopmental disorders and use this definition to i) review evidence for compensation, garnering examples from ASD, ii) propose a preliminary framework of the workings of compensation, iii) explore its potential cognitive and neural underpinnings, and finally, iv) discuss the research and clinical implications of studying neurodevelopmental disorders within this compensation framework.

2. Defining and measuring compensation in neurodevelopmental disorders

2.1. Compensation in the psychological literature

Within psychological research in general, the term 'compensation' has been widely used. In instances where participants, who are expected to be limited in a particular set of resources (be this due to a psychiatric condition, old age, or an experimental manipulation amongst healthy participants), perform better than expected on a psychological task, the possibility that they have in some way compensated, is often speculated upon by authors. This compensatory hypothesis is generally backed up by evidence showing that 'compensated' participants have achieved this 'typical' performance with the recruitment of additional resources, be these neurobiological, cognitive, or genetic. For example, in the literature on aging, researchers have used the term to describe how older adults can demonstrate atypical activation (enhanced or decreased) of task-relevant brain areas or activation of additional regions not typically recruited by younger adults, in order to perform a task just as well as their younger counterparts (see Grady, 2012). Equally, within research into neuropsychological patients, the term 'compensation' refers to the brain's ability to rely on alternative neural routes after typical routes have been compromised by brain damage, in order for patients to make improvements in behaviour/cognitive abilities (e.g., Price and Friston, 2002). The term 'partial compensation' is used to describe how an attempt to counteract limited resources may not always be efficient enough to support wholly 'typical' behaviour or cognitive task performance.

Despite frequent use of the term 'compensation', there is no technical or universal definition. The precise interpretation of its meaning or the meaning of 'compensatory brain activity' is specific to the particular task and participant population in question. The literature also suggests that the process of compensation could actually exist and operate at multiple levels, from molecular and/or genetic pathways (for instance, synaptic plasticity in order to counteract atypical connectivity; e.g., in ASD, Bourgeron, 2015) to broader cognitive systems and behaviour (for instance, atypical neural functioning to support typical cognitive task performance; e.g., in ASD, White et al., 2014). In this paper, due to the complexity and novelty of the phenomenon, we will focus on conceptualising compensation across levels of behaviour, cognition and whole neural networks only, solely within the scope of neurodevelopmental disorders.

2.2. Compensation in the literature on neurodevelopmental disorders

With no agreed definition, it is unsurprising that compensation in neurodevelopmental disorders has received little empirical attention. There is, to our knowledge, only one review paper on the topic (Ullman and Pullman, 2015), which explores the specific compensatory function of the declarative memory system in five neurodevelopmental disorders, including ASD. Crucially, the authors' review relies on a definition of compensation that is reminiscent of that described in the aging and neuropsychological literature; that compensation reflects how an intact neurocognitive process/system might take over, or *compensate for*, the functioning of a defective process/system in order to maintain typical behaviour and/or cognitive task performance. Indeed, Ullman and Pullman (2015) suggest that in ASD, where socio-cognitive functioning is compromised, intact declarative memory ability may scaffold social behaviour; for example, the ability to recall previously learned social rules may replace intuitive understanding of social cues, thereby contributing to an appropriate social response.

There is, however, good reason to question whether a definition derived from the study of individuals who have *acquired* their deficits (e.g., brain-damaged individuals/aging adults), necessarily extends to neurodevelopmental populations (Johnson, 2017; Thomas and Karmiloff-Smith, 2002). For example, Johnson (2017) has highlighted how focal brain damage during the pre/perinatal period may be compensated for by early reallocation of function to intact brain regions, but that in the case of conditions such as ASD, where more wide-spread early brain disturbance is observed (or postulated, e.g., general synapse dysfunction), an alternative explanation of compensation may be required. Additionally, brain injury in healthy adults may trigger a host of compensatory processes (e.g., enhanced connectivity from damaged to frontal regions; Sharp et al., 2014) that are not necessarily comparable to cases where a cascade of atypical neural function has existed from very early in development.

In our endeavor to find a definition of compensation drawn from observations in neurodevelopmental phenotypes, we take inspiration from research into a developmental condition that has a relatively circumscribed cognitive deficit and has received some preliminary discussion with regards to compensation; namely developmental dyslexia.

2.3. Lessons from developmental dyslexia

Developmental dyslexia is characterised by a specific impairment in reading, not otherwise accounted for by intellectual or visual abilities (American Psychiatric Association, 2013). The condition is proposed to be underpinned by a core deficit in phonological processing (Snowling, 2014), which contributes to an array of behavioural symptoms amongst dyslexics (e.g., spelling errors and slow reading and word recognition; Thambirajah, 2010). Critically, although the majority of children with dyslexia experience these difficulties persistently (Hatcher et al., 2002; Maughan et al., 2009), a subset of individuals, referred to as 'compensated dyslexics' (Lefly and Pennington, 1991), eventually establish typical reading skills by the time they enter adulthood (Callens et al., 2012; Gallagher et al., 1996).

In principle, there are at least three possible ways in which dyslexic individuals' primary symptoms could lessen. First, the phonological processing deficit at the cognitive level may genuinely remit, thus supporting good reading ability. Second, phonological processing may be delayed rather than deficient in these children, so that there is eventually developmental 'catch up'. Third, good reading ability may be facilitated by alternative neurocognitive pathways that are independent of phonological routes. On inspection of the literature, the first two possibilities do not appear to hold up empirically. Amongst highly 'compensated' dyslexics, who do not exhibit measurable spelling or reading difficulties (e.g., those in higher education), significant phonological processing deficits are revealed when tapped with sensitive enough cognitive probes, such as rapid picture naming (Gallagher et al., 1996; Ingvar et al., 2002; Parrila et al., 2007; Swanson, 2012). Further, these individuals' reading abilities are not necessarily comparable to those of typically developing individuals under certain