

[NARRATIVE REVIEW]

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Gravitational insecurity: A brief introduction to a condition of life-long balance challenges identified by occupational therapists

ABSTRACT

Background: Gravitational insecurity (Grl) involves life-long balance problems that impact activities of daily living like climbing in children and tilting back tasks in adults. It increases fall risk, as recently found. Understanding Grl pathophysiology is increasing but remains incomplete. Grl is unrecognized by the larger health provider community. Treatment options are effective for some clients but need expansion.

Methodology: This review describes the presentation, prevalence and co-morbidities of Grl in children and adults. Explanation of likely pathophysiological causes of Grl based on collaborative neuroscientific/occupational therapy research is presented in the context of vestibular system function. Rationales for future studies are explained. A new treatment modality based on current vestibular rehabilitation is suggested.

Findings: Children with gravitational insecurity comprise up to 5% of clients seen by many paediatric occupational therapists. About 2% of American adolescents and adults experience clinical Grl; another 17% have milder balance issues. Fine and gross motor difficulties and sensory hypersensitivities are common co-morbidities in children and adults, respectively. Grl may involve deficiencies in brainstem “vestibular velocity storage” that inadequately amplifies and prolongs signals of small, slow head rotations necessary for balance. Such deficiencies may be partially compensated by greater reliance on neck muscle proprioception. Galvanic vestibular stimulation (GVS) is a non-invasive treatment holding promise for Grl.

Conclusions and recommendations: Understanding Grl can be broadened by research in additional countries and by categorizing its heterogenous presentation. Understanding can be deepened by assessing the relationship of tilt intolerance to VVS anomalies. Research into GVS for Grl should be undertaken. Such research can inform the larger health provider community about Grl, much to the credit of OTs who identify and treat it.

Implications for practice: Understanding Grl vestibular pathophysiology is important for improving the outcomes of therapeutic interventions used to ameliorate the condition. Galvanic vestibular stimulation is a new treatment modality deserving of further research into its usefulness for Grl.

INTRODUCTION

Gravitational insecurity was first identified by A. Jean Ayres in her development of sensory integration typologies and interventions¹. Grl interferes with child and adult ADLs like climbing play structures in children and stepladders in adults and tilting back activities, like tumbling in children and hair-washing in adults². And, as recently discovered, Gravitational insecurity increases fall risk³. Although occupational therapists now recognize the condition, the physiological bases of Grl remains incompletely understood. This lack is important because a rational, empirical approach to intervention depends upon an understanding of the nature of the problem. May-Benson and Kumar⁴ conjectured about the possibility of vestibulo-cerebellar deficits but did not specify what they might be nor how they would account for the presence of balance problems and the absence of typical vestibular

signs like dizziness and vertigo. In the years since Ayres work a great deal of knowledge about vestibular function has been accumulated that is relevant but has not been applied to the understanding or treatment of GrI. Also and unfortunately, an interpretation of GrI as a "hypersensitivity to gravity" has been promulgated which is misleading and counter-productive to understanding its real physiological bases.

Therefore, this review focuses on our studies which have taken an evidence-based, collaborative neuroscientific/occupational therapy approach to determining the pathophysiological bases of GrI. The order of their presentation in this narrative review follows that of our progressively deeper research into the characteristics of GrI and the vestibular dysfunction that is causal to it. In what follows, we cover the presentation and prevalence of GrI in children and adults, co-morbidities that may accompany the condition but are not an integral part of it, relevant aspects of normal and pathophysiological vestibular function and, lastly, standard interventions as well as a potential new therapeutic modality.

The information presented in this narrative is important to readers of the South African Journal of Occupational Therapy in terms of identifying the condition of GrI, citing sources that provide deeper background into its characteristics and intervention, inviting those with clinical and/or research interests to contact the author for further discussion and, perhaps, inspiring exploration of GrI in Africa from which there have been no reports on it.

LITERATURE REVIEW

The literature on GrI is relatively limited. May-Benson's² scoping review of GrI in both children and adults involved an extensive search for research studies, review articles and book chapters written in English and published between 1972 and 2016, as well as grey literature (e.g., theses, dissertations, posters, unpublished papers) from this period. Her search yielded only 22 documents, 18 of which were generated by three authors/laboratories (i.e., Koomar, May-Benson or Ganapathy Sankar) in the US or India. Most of these dealt with assessment. The only work on physiology not otherwise cited in this narrative is Weisberg's observation of several children with a high resting state of arousal as measured by skin conductance (i.e., sweating) who demonstrated fearful responses to unexpected movements⁵. A subsequent Scholar Google search for papers published since 2016 found four: Matson et. al.⁶ and Pierce et. al.⁷ on perceptions of and educating about GrI, Watkyns et. al.⁸ on assessment and Desale et. al.⁹ on intervention. It is highly unlikely that peer-reviewed, neuroscientifically adequate research by established researchers on the pathophysiology of GrI would not be found on Scholar Google.

The intent of this narrative is to cover what has been discovered about GrI pathophysiology and its implications and applications. As noted, this narrative does review crucial basic elements of vestibular anatomy and physiology as well as recent findings about vestibular function and dysfunction in the biomedical literature as they pertain to GrI as background. Information about GrI itself is drawn from two studies published by Potegal and colleagues, a third study under review and a fourth study in progress, as indicated in the respective sections of the text that follows. We intend this narrative to be useful to readers who might wish to explore that literature. In this context we note the term "vestibular hypofunction" as used in that literature refers to a reduction in, or absence of, signals from the semicircular canals and/or the otolith organs themselves or in their ascending neural pathways, which results in balance problems and associated issues.

FINDINGS

Presentation and prevalence in children

GrI presents in childhood as a reluctance to move the head out of an upright orientation (e.g., to lay back or somersault), sensitivity to motion (e.g., becoming car sick) and fear of losing balance (e.g., on uneven or yielding surfaces or playground equipment²). These children often refuse to climb, swing, skate or bike. They prefer sedentary activities and may be teased about their clumsiness and/or non-participation in sports.

Clinical identification of GrI in children often occurs through a series of structured observations¹⁰. Standardized measures of sensory processing, such as Sensory Processing Measure-2 also contain items that may help identify GrI¹¹. May-Benson & Koomar's validated behavioral tests for identifying GrI in children include rising from supine lying on therapy balls and stepping on/off tiltboards⁴. These measures for GrI identification in American 5-10 year olds were cross-culturally confirmed in Indian 3-9 year olds¹². Unfortunately, these behavioral tests are under-utilized by practitioners¹³.

There are no data on the prevalence of GrI in children at large in any country. Up to 5% of child clients seen in pediatric occupational therapy clinics in the US are identified with GrI, but this increases to 11-20% of the children seen by occupational therapists in private practice^{2,13}. Girls may be more frequently affected than boys, which is the reverse of the general tendency for more boys to be seen in these clinics, but is consistent with increased female vulnerability to balance issues and vestibular dysfunction, at least in adults¹⁴.

Presentation and prevalence in adults

GrI in adults presents as a hesitation about, or avoidance of postures and movement activities that challenge balance, such as standing on one foot². These adults are alert to and cautious about standing or walking on substrates that are uneven, soft or yielding, or tilted. They may avoid tilting back in activities such as reaching for a high shelf, leaning back in a chair or getting their hair washed at the salon. They are wary of heights, like standing on a stepladder; reports of particular difficulty managing transport on escalators are frequent and consistent.

People with GrI often describe themselves as "always wanting their feet on the ground". Through our interaction with these individuals we came to realize that they meant this literally, not metaphorically, suggesting that they may be more dependent on pressure receptors in the soles of the feet and proprioceptors in leg muscles for balance. Objective dynamic posturographic analysis showed that, when challenged by reduction of relevant optic flow and ankle proprioceptive cues, people with GrI were able to maintain the same level of standing balance (measured by body sway) as did healthy subjects by switching from a less effortful ankle based strategy of postural control to a more effortful hip strategy¹⁵. This particular observation is consistent with the general impression that people with milder forms of GrI with fewer and/or less severe symptoms struggle more-or-less successfully with their balance issues. However, recent evidence indicates that GrI does increase fall risk, making this condition a more serious health problem than previously recognized².

People with GrI experience distinct but heterogeneous patterns of difficulty². During interviews early in our research program several individuals revealed problems with visual-spatial skills and spatial orientation, e.g., difficulties while driving, frequently getting lost in new environments. Formal follow-ups with a standardized questionnaire confirmed spatial disorientation as a consistent characteristic of GrI. This issue is likely due to anomalies in the vestibular signals ascending to parietal cortex where they are incorporated into representations of spatial location and direction that guide way-finding. Contrary-wise, people with GrI do not complain about classic vestibular signs of dizziness, vertigo or oscillopsia (illusory movements¹⁶). This is important because these signs are more typical of dysfunction in the inner ear or vestibulo-cochlear cranial nerve VIII, again suggesting that the problems of GrI exist at more rostral levels of the nervous system. Accordingly, dizziness and vertigo have been exclusion criteria in recruitment of participants in our GrI research.

GrI and the difficulties with activities of daily living ADLs associated with it can lead to uncomfortable subjective experiences and self-evaluations. Some people depend on vision to feel grounded and experience anxiety-provoking disorientation when their eyes are closed. One research participant asked: "Am I just clumsy or really crazy?" She was assured that she was not crazy. In our experience, individuals with GrI are relieved to be told that their experiences are not idiosyncratic or

weird but they, like others, have an understandable and possibly remediable condition.

A survey of 1991 adolescents and adults estimated that GrI affects 2% of the US population at large; another 17% may have a mild form of the condition (i.e., fewer and/or less severe symptoms)². Females had more severe symptoms in keeping with other observations of female vulnerability to these issues¹⁴.

GrI CO-MORBIDITIES

GrI co-morbidities in children

Children with GrI typically present in occupational therapy clinics with a number of co-morbidities, often including core weakness and fine and gross motor difficulties that, conjecturally, might qualify for a diagnosis of Developmental Coordination Disorder. Although the balance problems that define GrI are rather consistent from child to child, they could just result from these multiple co-morbidities. This question of whether GrI is really a definite condition by itself was provisionally resolved in a survey of pediatric occupational therapists in which an estimated 6% of children in their clinics presented with the balance issues of GrI but without the motor difficulties¹³. This resolution was subsequently strengthened by informal but extensive interviews with adults about their GrI experiences as well as by the research presented below.

GrI co-morbidities in adults

Adult occupational therapy clients with GrI also present with a number of co-morbidities, often including multiple sensory hypersensitivities. These aversive reactions to lights, sounds, touches and tastes have been understandably but unfortunately over-generalized to the hypothesis that GrI represents a "hypersensitivity to gravity" (e.g.,¹⁷), a force that, unlike sensory stimuli, is continually and unvaryingly present and for which there is no biophysically plausible model of hypersensitivity. Indeed, no correlation was found between sensory hypersensitivity and an index of GrI severity, a result which has been recently replicated^{3,15}. GrI more plausibly involves anomalies in the central processing of signals from semicircular canals and otolith organs, as described below.

Anxiety also appears as co-morbid with GrI in some children and adults. Although there is evidence that anxious people are more prone to balance problems and vice versa¹⁸, anxiety does not seem to be an intrinsic feature of GrI^{15,19}, just as it is not associated with even more severe vestibular hypofunction²⁰. Nonetheless, people with GrI who do experience feelings of anxiety may be misdiagnosed as having mental health disorders by medical and psychological practitioners unfamiliar with GrI.

NORMAL VESTIBULAR FUNCTION

Lower-level reflex stabilization

Functionally, the vestibular system operates at several levels. The lower, brainstem levels provide reflex stabilization of gaze and posture; the higher, cortical levels contribute to the conscious perception of self-motion, navigation through the spatial environment and other cognitive functions. Anatomically, signals driving all these functions originate from the semicircular canals and otolith organs of the inner ear that lie within a complex hollow chamber in the petrous bone of the skull. The three fluid-filled semicircular canals detect head rotations in the horizontal (yaw), vertical (pitch) and lateral (roll) planes. They do so because head rotation bends the cupula, a little flap inside each canal. When the cupula bends, signals from the sensory hair cells embedded in it drive the vestibulo-ocular reflex (VOR), which is compensatory eye movement opposite to the head rotation that stabilizes the direction of gaze. When we spin around a few times and then stop, the VOR remains active for a while, driving the eye movements of post-rotary nystagmus (PRN) which we perceive as the visual field repeatedly flicking in one direction. PRN happens in part because it takes some seconds for the cupula to unbend back toward its normal, unbent position.

Bending of hair cells embedded in the otolith organs (sacculae and utricle) detects both the orientation of the head to gravity, as in head tilt,

and linear acceleration of the head, e.g., as in walking forward. Otolith organ signals drive spinal reflexes that stabilize upright posture. However, because their hair cells detect both head tilt and linear acceleration, it is crucial that their signal be disambiguated so that the brain receives accurate information about which type of movement is happening. It is signals from the canals that disambiguate the otolith organ input: head tilt involves a rotation that is signaled by the canals but linear acceleration does not activate canal hair cells.

However, there is an additional complication. While the canals reliably detect the larger, more rapid head movements involved in looking around, their biophysics limits their ability to transduce smaller, slower movements that happen all the time. For this reason, we and other animals have evolved a vestibular velocity storage (VVS) circuit in the brain that amplifies and prolongs these small, slow canal signals ("gain" is the technical term for amplification). The VVS circuit in our brains has been recently localized to cerebellum and vestibular nuclei of the brainstem²¹. A telltale sign of signal prolongation by the VVS circuit is that while the cupula returns to its normal, unbent position within about 5 seconds after spinning, PRN usually lasts around 15 seconds. The additional 10 or so seconds is totally due to activity in the VVS. This duration of activity, controlled by the "time constant" of the VVS, is critical to balance. It needs to be long enough to insure accurate VOR function, but if it were any longer it would be susceptible to noise that would infiltrate and degrade the signal²². Shorter than normal VVS time constants have been found in fall-prone older individuals²³ while excessively prolonged time constants are associated with motion sickness, migrainous vertigo and Meniere's disease²⁴⁻²⁶.

Higher level vestibular functions

Projections from the vestibular nuclei ascend to at least 5 cortical areas. Input to ventral temporo-occipital regions subserves conscious sensations of self-movement²⁷. Vestibular input to parietal cortex contributes to distance judgments and other aspects of spatial processing and navigation²⁸⁻³⁰. Input to other cortical areas is involved in various cognitive functions beyond the scope of this narrative³¹.

GrI PATHOPHYSIOLOGY

Occupational therapist's, observations of children with GrI

Respondents to the Paediatric Occupational Therapy Survey¹³ of GrI endorsed significant overlap of GrI presentation with classic signs of vestibular dysfunction like unsteady balance, broad-based gait, and eye movement abnormalities¹⁶. A lack of overlap with indicators of more common childhood diseases suggested that the overlap with vestibular signs was not due to a negative halo of perceived general symptom-proneness. Indeed, Ayres had found shortened, or sometimes prolonged, PRN in children with GrI, which she conjectured might involve anomalies in some vestibulo-cerebellar neurocircuitry³². The VVS is most likely to be the circuitry in question. Rigorous testing of the VVS hypothesis requires standard, vestibular clinic diagnostics that involve rotation in the dark. Children are not suitable subjects for such tests, but adults with GrI are.

Studies of adults with GrI

Adults with life-long GrI and sex, age and ethnicity matched comparison subjects without GrI were identified by a reliable and valid 8 item screener of recalled childhood balance experiences¹⁵. Rotation at the slowest frequency revealed a combination of reduced gain and shorter time constants (i.e., shorter nystagmus durations) that together correlated with GrI severity. There were no between-group differences at higher rotation frequencies for which VVS is not needed and does not operate. These results closely confirm and extend earlier work by Fisher et al³³. There was also a trend for increased gain at the higher rotation frequencies, perhaps representing some sort of compensatory adaptation. Thus, less robust and/or anomalous VVS signals may well be part of the cause of the movement/balance problems in GrI. Repeated findings of co-occurring spatial disorientation and problematic way-finding, assessed with the standardized Santa Barbara

Sense of Direction Scale³⁴, might hypothetically result from anomalous VVS signals ascending via known vestibular projections to cortex where they are incorporated into spatial navigation circuitry^{30,35}.

In any case, these studies left open the possibility that GrI might also involve deficiencies in signals of head orientation to gravity originating from the otolith organs, as suggested by Ayres and Tickle³⁶. This hypothesis was tested in a follow-up study of the subjective visual vertical (SVV) in which people sitting in a chair set a pointer to what they perceived to be the upright direction (i.e., the 12 o'clock position) in an apparatus that eliminated external visual cues³. Setting the SVV is now accepted as a measure of otolith organ function, both experimentally and clinically, e.g., in evaluating impairment, recovery and treatment effects for a range of conditions affecting vestibular function and balance in loci from inner ear to cortex^{37,38}. Because neck muscle proprioception also contributes to head orientation in general and to SVV judgments in particular, brief, high frequency neck muscle vibration was used to assess proprioceptive contributions to verticality settings, per³⁹.

The primary findings were that vibration had significantly greater effects on verticality settings in the GrI group and that the magnitude of the vibration effects increased with the severity of GrI. These results suggest that people with GrI may rely more on neck muscle proprioception to monitor head orientation, perhaps because their otolith organ input is less reliable. There were no group differences in baseline, non-vibration conditions, which may have been due to the successful substitution of neck proprioception for otolith organ input in the non-challenging situation of making verticality judgments while seated. However, relying on neck muscle proprioception rather than otolith organ input for monitoring head orientation would be problematic under more challenging, real life balance conditions because neck muscle proprioception is less accurate than otolith organ in head tilt detection⁴⁰.

Overall, the evidence base to date suggests that the balance problems of GrI may originate in inadequate VVS amplification and prolongation of small, slow head rotations signalled by the semi-circular canals. This deficiency may be partially compensated by a greater reliance on neck muscle proprioception, which is less accurate than proper otolith organ input. GrI has sometimes been ascribed to a supposed hypersensitivity to gravity¹⁴, an hypothesis which has no known basis in physics and is likely a conflation of co-morbid but uncorrelated sensory hypersensitivities compounded with the emotional distress of experiencing imbalance. Greater attention to the demonstrable pathophysiology of imbalance is warranted.

Hypotheses about co-morbid sensory hypersensitivities

But, what of the co-morbid sensory hypersensitivities of adults with more severe GrI who appear in occupational therapy clinics? It may be that individuals with multiple issues are more likely to seek professional help than those with mild challenges, so present with a larger range of more serious problems. Additionally or alternatively, the VVS anomalies that cause GrI and the frequently co-occurring morbidities might arise from functionally unrelated processes. For example, the vestibulo-cerebellar circuitry of VVS is replete with B type receptors for the powerful inhibitory neurotransmitter GABA⁴¹. The shortening of the VVS time constant by the GABA-B agonist baclofen (Lioresal) confirms the role of these receptors in balance⁴². But, GABA-B receptors are also found in other sensory systems, e.g., their modulation of olfactory and auditory processing has been demonstrated pre-clinically, while GABA itself plays a role in tactile processing in human thalamus and sensorimotor cortex⁴³⁻⁴⁶. So, generally malfunctioning GABA-B receptors might well create vestibular dysfunction that correlates with an assortment of otherwise unrelated behavioural problems. That is, in this example the sensory hypersensitivities of individuals with more severe forms of GrI may occur because GABA-B receptors happen to play a role in a number of functionally independent neural circuits.

FUTURE RESEARCH STUDIES

Up to now, GrI has been recognized almost exclusively by occupational

therapists. It is largely unknown to the greater health provider community. To bring GrI to the attention of physicians, psychologists and others who are likely to misinterpret the balance complaints of their patients, more solid evidence about its physiological bases would be instrumental. The broader recognition of GrI would also redound to the credit of occupational therapists who have identified an otherwise unidentified form of mild, idiopathic, bilateral vestibulopathy. Some research to advance this effort is currently in the planning stage.

Do tilt tolerance thresholds correlate with reductions in VVS time constant?

In the Paediatric Occupational Therapy Survey¹³, 65% of respondents reported children's problems with lying supine; only 17% reported problems with lying prone. Backward tilt intolerance in adults is more anecdotal and has not been systematically measured but could potentially provide a quantitative index of GrI severity. Furthermore and importantly, it might be quite directly related to the VVS time constant. Upward head pitch or supine lying reduces the VVS time constant in cats, monkeys and humans⁴⁷⁻⁴⁹, but increases the gain of supra-threshold rotational signals as measured by vestibulo-ocular and optokinetic nystagmus in animals and people⁵⁰⁻⁵². Thus, the aversion of people with GrI for tilting backward could be explained if it further, and perhaps disproportionately, reduced their VVS time constant, which is already too short, and/or increased their suprathreshold gain (e.g., overestimate of head tilt), which may be already too great. We are preparing to test the hypotheses that people with GrI will reach a threshold of discomfort with lesser backward tilt and that this tilt tolerance threshold will correlate with a disproportionate, tilt-induced reduction in VVS time constant.

Vestibular diagnostic studies of otolith organ function

Newly standard clinical diagnostic tests of otolith organ function include vestibular evoked myogenic potentials (VEMPs) in which loud auditory clicks stimulate hair cells of the saccule and utricle and the resulting contractions of ocular or neck muscles are recorded by EMG⁵³. By varying the loudness of the clicks, VEMP testing could determine if individuals with GrI have higher thresholds for activating the otolith organs then matched comparison subjects.

GrI heterogeneity

Patterns of GrI presentation appear heterogeneous. There are people who tend to bump into things and describe themselves as "clumsy", others who have particular difficulty with heights and on escalators, and those who become motion sick. Some individuals with GrI also report unusual, movement-related perceptual experiences such as occasional, unprompted sensations of dropping or feeling overwhelmed by the onrush of the visual scene as they walk forward. Such experiences might reflect anomalous signals ascending to one or more areas of cortex that process vestibular information. Given that different kinds of interventions might work best for people with different sorts of experiences, a combination of open-ended interviews and broad questionnaires could be used to acquire and sort GrI-related experiences into nosological categories using factor analysis, latent class analysis or related statistical procedures.

National studies

There has been no work on GrI in Africa, to the best of our knowledge. Because ethnic differences in balance and vestibular function do exist⁵⁴, determining the prevalence and characteristics of GrI among groups in Africa would be a welcome addition to knowledge.

INTERVENTIONS FOR GrI

Current practice

Pediatric Occupational Therapists with advanced training in sensory integration and processing treat GrI with child-centered sensory-motor experiences designed to challenge and enhance motor, postural, and praxis skills^{11,55}. These interventions most commonly include play-based head movements out of the midline and body position changes along

with calming sensory input, i.e., proprioceptive and deep pressure tactile stimulation⁵⁵. Occupational therapists variously report that these interventions reduce fear and increase participation in activities such as swinging, climbing, and ADLs. However, systematic peer-reviewed evidence on treatment effectiveness remains limited⁵⁶.

Respondents in Potegal et al's¹³ Paediatric Occupational Therapy Survey reported that they had first introduced gentle linear movement, then gradually moved to rotation exercises with 64% of child clients and used Astronaut Training⁵⁷ with 38%. Roughly a third of the children were reported to show little improvement or actually got worse, about a third became somewhat more functional while a third reached a point of experiencing unrestricted movement. Some in the latter group began to "crave" movement and ask for experiences that they had previously feared and avoided. While the latter results are encouraging, overall, these outcomes are consistent with the generic "rule of thirds" for treatments of a variety of medical conditions (e.g.,⁵⁸). There are no reports of outcomes for adults with GrI. Surely, it is possible to do better.

Galvanic vestibular stimulation

Within the rapidly developing field of vestibular rehabilitation, galvanic vestibular stimulation (GVS) has been reported to improve balance function and/or gait in healthy older adults and in patients with Ménière's disease, vestibular neuritis/schwannoma, vestibular hypofunction, Parkinson's disease and stroke^{59,60}. GVS involves placing non-invasive electrodes over the mastoid processes and passing low currents through vestibular system of the inner ear for 5 – 30 minutes. Among the various GVS stimulation protocols in use, subthreshold, random-impulse ("noisy") GVS that creates "stochastic resonance" in which naturally occurring, but below-threshold vestibular signals are boosted into a normal range is of particular interest⁶¹. This GVS mode has advantages in having no adverse side-effects and, imperceptible to participants, easily permits placebo control trials required for rigorous treatment evaluation. GVS is most effective in those with at least some vestibular function, which people with GrI surely have. The therapeutic possibilities of galvanic vestibular stimulation for GrI are quite promising.

A final word

This paper has been written with the aim of encouraging further research on GrI. We welcome contact with readers who might be interested in these issues.

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