Medical Hypotheses 80 (2013) 234–236

Contents lists available at SciVerse ScienceDirect

Medical Hypotheses

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

Is abnormal vestibulomotor responses related to idiopathic scoliosis onset or severity?

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ARTICLE INFO

Article history:
Received 20 July 2012
Accepted 25 November 2012

ABSTRACT

Results from several studies have suggested that brainstem dysfunction occurs more often in adolescent with idiopathic scoliosis compared to healthy individuals. The vestibular nuclei occupy a prominent position in the brainstem. Because the lateral vestibulospinal tract controls axial muscles, alteration in the brainstem during body growth (i.e., preadolescent and adolescent period) may translate in abnormal trunk activation and thus cause permanent spinal deformities. We conceive that vestibular dysfunction may be observed only in AIS patients with severe spine deviation. Consequently, adolescent with severe idiopathic scoliosis (AIS) would exhibit abnormal vestibulomotor responses compared to healthy age-matched individuals and AIS patients with mild spine deformation. If this hypothesis is confirmed, it will suggest that abnormal vestibulomotor response may contribute to curve progression. On the other hand, if AIS patients with mild severity also show abnormal vestibulomotor response, it will indicate that impaired vestibulomotor may be related to scoliosis onset but is not necessarily related to curve progression. It is expected, however, that regardless of curve severity, not all patients would have abnormal vestibulomotor responses. For instance, in some cases, gene defects may lead to malformation of the semicircular canals or alteration of the vestibular cortical network and cause scoliosis or curve progression.

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Introduction

Adolescent idiopathic scoliosis (AIS) is a three-dimensional structural deformity of the spine. It is predominant in females with a gender ratio ranging from 5.4:1.0 to 23.9:1 for curve greater than 20° and this ratio increase with curve severity and age. It has a prevalence of 2–3% and an annual incidence of 4.5% [1–3]. Although the etiopathology of AIS remains unclear, it has been proposed that its causes are multifactorial: genetic factors, neurological mechanism dysfunctions, hormone/metabolic dysfunctions, skeletal growth abnormalities, and biochemical factors [4–6]. Among all the factors that may be related to scoliosis, there are several evidences that some neurological mechanisms are impaired in patients with AIS. One structure that has attracted attention is the brainstem. For instance, Yamamoto et al. [7] have observed a positive correlation between brainstem dysfunction, determined by visual target pursuit tests, and curve progression. The authors concluded that brainstem alteration might be related to curve progression. Another oculomotor disorder, lateral gaze palsy, is associated with a high prevalence of idiopathic scoliosis. This observation suggests that the paramedian pontine reticular formation would be impaired in adolescent idiopathic scoliosis patients [8]. Furthermore, by investigating postrotatory nystagmus, other researchers have also concluded that patients with AIS had brainstem dysfunction [9,10].

The vestibular nuclei occupy a prominent position in the brainstem. Because the lateral vestibulospinal tract controls axial muscles, alteration in the brainstem during body growth (i.e., preadolescent and adolescent period) may translate in abnormal trunk activation and cause permanent spine deformities. Consequently, during the last decades, several authors have suggested that scoliosis onset may be related to impairment of the vestibular system because of its contribution to regulation of axial muscles tone e.g. [5,11–13]. Nonetheless, no study has addressed whether direct vestibular stimulation would lead to abnormal motor response in patients with AIS compared to controls. The following sections explore in detail each point involved in the formulation of the hypothesis.

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http://dx.doi.org/10.1016/j.mehy.2012.11.035

The nervous system

Approximately three decades ago, it has been proposed that AIS could be associated to neurological impairments [14]. Consequently, balance control of patients with AIS has been extensively studied as it requires proper integration of visual, proprioceptive and vestibular and also appropriate sensorimotor transformations to generate balance motor commands that reduce body sways [15–18]. Results from studies that assessed balance control of patients with AIS have shown that sensory deprivation or transient sensory reintegration induce a greater increase in body sway in patients with AIS compared to control group participants [19–22], supporting the suggestion that scoliosis is associated to abnormal sensory processing in the cerebral cortex. This suggestion is corroborated by results from various studies that have found abnormal somatosensory evoked potential in patients with AIS [23–26]. Furthermore, this suggestion is also corroborated by the fact that patients with AIS have cognitive vestibular integration impairment [12], emphasizing the idea that some cortical mechanisms might be involved in scoliosis onset or curve progression [14,27]. Overall, these observations support the suggestion that alteration in the transformation of sensory information in motor commands may create asymmetrical axial muscle activities and develop in scoliosis.

The vestibular system

The vestibular system is essential in maintaining balance through vestibulospinal reflexes. Abnormalities in this system could thus induce faulty sensory inputs to the CNS and lead to inappropriate phasic and tonic trunk muscle activities. As mentioned above, such an altered motor drive could represent a predisposition to spinal deformity, mainly during the development of the spine throughout growth. As a matter of fact, some studies have shown malformations of the inner ear structures in patients with AIS: semicircular canals anomalies, lateral canal aplasia, and inadequate communication between the lateral and posterior semicircular canals. For instance, Shi et al. [28] observed a decrease in the distance between centers of lateral and superior canals in the left-side vestibular system. Results from another study have confirmed morphological abnormalities in patients with AIS with small curve deformity [13]. Overall, these results suggest that spine deformity may occur due to malformation of the vestibular apparatus. Results from these studies, although important, do not inform whether abnormal vestibular morphology is related to scoliosis severity. Furthermore, it is unclear if abnormal vestibulomotor response could occur in absence of abnormal vestibular morphology. In a previous study, we assessed, in patients with AIS presenting severe spine deformity (i.e., Cobb's angle > 28°) and controls, their capacity to integrate vestibular information for cognitive processing for space perception. Seated participants experienced rotations of different directions and amplitudes in the dark and produced saccades that would reproduce their perceived spatial characteristics of the rotations [12]. Compared to healthy controls, patients with AIS underestimated the amplitude of the rotations to a greater extent. It was suggested that severe spine deformation developed because of impaired vestibular information travelling from the cerebellum to the vestibular cortical network or due to alteration in the cortical mechanisms processing the vestibular signals. It is worth noting that the vestibulocerebellum receives information from both the semicircular canals and the otolith and that the Purkinje neurons in the vestibulocerebellum inhibit neurons in the lateral vestibular nuclei. Therefore, it is possible that impairment in the vestibulocerebellum could be related to scoliosis severity. This suggestion is supported by the proposition that deficit in melatonin suppresses the activity of the vestibulospinal tract, leading to abnormal muscle activities of the cervical and axial muscles [5].

The hypothesis

Based upon these observations, it is hypothesized that patients with AIS presenting severe spine deformity (i.e., Cobb's angle > 35°), compared to healthy control subjects and patients with moderate AIS (i.e., Cobb's angle between 20° and 30°), would exhibit larger vestibulomotor responses. If this hypothesis is confirmed, it would suggest that abnormal vestibular processing might be involved in curve progression. In contrast, if patients with moderate AIS also have abnormal vestibulomotor responses, it would suggest that scoliosis onset, but not necessarily curve progression, is related to impaired transformation of vestibular signals into proper motor commands. Moreover, we will assess if abnormal vestibulomotor responses is related to impairment in the development of the vestibular apparatus or if it is caused by sensorimotor transformation.

Simplifying the aetiology of AIS to a single factor would not be appropriate and one must recognize that some AIS patients with severe (or mild) spine deformity may not show abnormal vestibulomotor response, which would mean that curve severity or scoliosis onset is mainly related to other factors (i.e., hormonal, gene defects not related to vestibular morphology). For instance, it is possible that vestibulomotor impairment result from genetic predisposition leading to either a malformation of the semicircular canals [29] or a dysfunction of the sensorimotor mechanisms involved in axial muscle tone regulation [5,12,27].

Evaluation of the hypothesis

The proposed hypothesis will be tested using bipolar binaural galvanic vestibular stimulation (GVS) in controls and patients with AIS of mild or severe spine deformity. Whole-body displacements (i.e., torso and pelvis kinematics) along the frontal plane as well as changes in vertical forces (i.e., loading/unloading) following GVS onset will be measured. Galvanic vestibular stimulation is an attractive tool to study the vestibular apparatus and the balance control system because it delivers a direct disturbance at the receptor level without stimulating other sensory channels. At least immediately after GVS onset, before other sensors (e.g., proprioception) are involved, it reveals the operation of the balance system to direct vestibular perturbation. The hypothesis would be confirmed if patients with severe AIS demonstrate a larger or smaller vestibulomotor response compared to age-matched healthy individuals. Furthermore, if patients with AIS of mild severity also have abnormal vestibulomotor response (i.e., larger or smaller torso and pelvis displacements), it would indicate that impaired vestibulomotor response is related to scoliosis onset but not necessarily to curve progression. Furthermore, using computational technique allowing segmentation of the vestibular system from high-resolution T2-weighted MR images, the geometrical development will be assessed. Therefore, it will be possible to verify if AIS patients with abnormal vestibulomotor responses have normal vestibular morphology.

Consequences of the hypothesis

If only AIS patients with severe spine deviation show abnormal vestibulomotor responses, it might be appropriate to assess vestibulomotor responses in order to screen AIS patients, as it would suggest that impaired vestibulomotor responses could be associated with curve progression. In addition, the verification of these

hypotheses will likely help genetic researches to target the genes that may be associated with curve progression. For instance, if abnormal vestibulomotor responses are observed and are caused by malformation of the vestibular apparatus rather than impairment in the transformation of the vestibular signal in motor commands modulating axial muscle activities, specific genes related to bone development could be targeted.

Conclusion

It is well recognized that the sensorimotor mechanism of AIS patients diverges from age-matched healthy individuals. Impairment in vestibular signal transformation into motor commands may lead to abnormal axial muscle activities and may generate spinal curvatures during growth. This observation could be related to gene defects related or not to the development of the vestibular apparatus or other biological alterations [29]. Nonetheless, a validation of the suggestion that scoliosis onset and curve progression could be the expression of a subclinical vestibulomotor disorder needs to be assessed. Finally, it is crucial to determine whether it is due to impairment in the processing of the vestibular signal at the cortical level or alteration in the development of the vestibular apparatus.

Conflicts of interest statement

None declared.

Acknowledgements

M.S. is founded by *Fondation Cotrel-Institut de France*, and J.P.P. has received a doctoral scholarship from *Fond de Recherche en Santé du Québec*.

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